Survivorship Care Preferences

A pilot study of young adults with cancer

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BACKGROUND: Adolescents and young adults (AYAs) face increased risk for physical, social, and cognitive late complications of cancer therapy. Strategies are needed to better engage AYAs in survivorship research.

OBJECTIVES: This study aimed to determine the feasibility of enrolling AYAs within six months of diagnosis to a survivorship study and assess their health concerns and survivorship care preferences.

METHODS: Patients aged 15–34 years who were diagnosed with leukemia, lymphoma, or sarcoma at three Connecticut hospitals from 2008–2011 were identified and recruited by mail. Participants and their physicians received a survivorship care plan. Participants completed surveys at 6 months and 18 months after diagnosis.

FINDINGS: Recruiting AYAs to survivorship research remains challenging, even when approaching them soon after diagnosis. Novel strategies are needed for nurses and other healthcare team members to engage AYAs in survivorship care and research.

ATTEMPTS HAVE BEEN MADE TO RAISE AWARENESS of adolescent and young adult (AYA) patients aged 15–39 years diagnosed with cancer because of their unique health issues (Smith et al., 2016). AYA patients experience cancer-related health disparities in terms of lagging survival rates for common cancers (Kent et al., 2015; Smith et al., 2016), underrepresentation on therapeutic trials (Freyer & Seibel, 2015; Harlan et al., 2011), and heightened risk for comorbid conditions (Nass et al., 2015). The survivorship period, after therapy ends, brings new challenges because of the late complications caused by previous curative chemotherapy and radiation therapy. These include infertility (Cardonick, 2016), second cancers (Nass et al., 2015; Smith et al., 2016), cardiovascular disease (Chao et al., 2016), musculoskeletal problems (Daniel et al., 2015), psychosocial distress (Tai et al., 2014), and cognitive difficulties (John, Sender, & Bota, 2016). Because AYA survivors are at a particularly critical juncture in their lives, impairment of cognitive function and medical interruptions can have devastating effects on these patients’ education (Parsons et al., 2012; Patterson, McDonald, Zebrack, & Medlow, 2015; Tai et al., 2012), attainment of employment (Parsons et al., 2012; Patterson et al., 2015; Tai et al., 2012), and ability to start a family (Bellizzi et al., 2012; Smith et al., 2016). Although AYA survivors deserve focused attention, this group is challenging to recruit and retain in research because of members’ geographic mobility, transition from pediatric to adult specialty and primary care providers, and hesitancy to have regular contact with the healthcare system because of under-insurance and out-of-pocket costs (Clinton-McHarg, Carey, Sanson-Fisher, & Tracey, 2011).

Participation rates in research studies among AYAs are notoriously low, with rates as low as 1.5% (Rabin, Horowitz, & Marcus, 2013) and most under 50% (Clinton-McHarg et al., 2011; Harlan et al., 2011; Hendricks-Ferguson et al., 2013; Link et al., 2009). Only one study was identified with a participation rate of 61%, but this was after multiple mailings and telephone calls that added up to a substantial recruitment cost per patient that could be cost prohibitive for many studies (Richards et al., 2010). Low rates of AYA participation in research have been attributed, in part, to loss to follow-up and to challenges connecting with socially and geographically mobile patients (Harlan et al., 2011; Nass et al., 2015). Historically, recruitment of AYAs has been conducted one or more years...
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“Participation rates in research studies among adolescents and young adults are notoriously low.”

After treatment completion, when AYAs are likely to be less connected to the oncology team and, consequently, harder to reach as well as less receptive (Clinton-McHarg et al., 2011; Gorman et al., 2014; Harlan et al., 2011; Rabin et al., 2013). Earlier recruitment, while patients remain under the care of a trusted oncology provider and are still connected to a particular system, could help to involve AYAs in survivorship research and care.

Psychosocial factors may also contribute to lack of engagement in research. The AYA period is often marked by upheaval and psychological distress, regardless of physical health status. The cancer experience places AYA survivors at heightened risk for the onset of serious mental health problems, including anxiety, mood, and substance abuse disorders (Daniel et al., 2015; McGorry, Purcell, Goldstone, & Amminger, 2011). Health concerns, psychological distress, and emotional and behavioral factors have been studied with regard to treatment adherence (Robertson, Wakefield, Marshall, & Sansom-Daly, 2015) and, to a lesser extent, research studies (Barnett et al., 2016). Because the diagnosis and treatment of cancer can create a psychological burden for this vulnerable population (Barnett et al., 2016), this may affect decision making regarding participation in survivorship research (Buchanan, Block, Smith, & Tai, 2014) and, consequently, be a point of possible intervention to improve AYAs’ participation.

The Health and Medicine Division of the National Academies of Sciences, Engineering, and Medicine (Institute of Medicine [IOM], 2013), the Livestrong Foundation, and the National Cancer Institute ([NCI], 2006) have issued statements and formal reports addressing the need to separate the ways in which cancers affect AYAs differently than other age groups (Nass et al., 2015; Smith et al., 2016). Care should be exercised in extrapolating data from AYA survivors of childhood cancers to patients diagnosed as AYAs, because these two groups are fundamentally different (IOM, 2013). Addressing health disparities experienced by AYAs will only be possible when effective strategies for their engagement in research have been developed and best practices for delivering survivorship care have been identified. Survivorship care seeks to improve long-term outcomes in patients with cancer through supportive care, prevention and intervention strategies to reduce the severity of late complications, surveillance in the high-risk patient subgroup, and health education (Denlinger et al., 2014). Individuals are considered survivors and are eligible for different aspects of survivorship care from the time of diagnosis through the rest of their lives (NCI, 2017).

The primary aim of this study was to determine the feasibility of recruiting and retaining AYAs within six months of their diagnosis to a survivorship study as a strategy to increase participation. The secondary aim was to describe and compare participant health concerns, psychological distress, and follow-up care preferences at enrollment (within 6 months of diagnosis) and at follow-up (18 months postdiagnosis). These three factors are thought to affect participation and recruitment into research and were pursued as a means to identify intervention targets and inform future strategies to heighten AYAs’ engagement in survivorship research and follow-up care.

Methods
This pilot cohort study of AYAs used the Connecticut Tumor Registry and the Rapid Case Ascertainment (RCA) Shared Resource of the Yale Cancer Center to identify potentially eligible AYA patients diagnosed within the previous six months from January 2008 to December 2011 at three large Connecticut hospitals. Additional eligibility criteria included the following: (a) a histologically confirmed malignant cancer typically seen in AYAs, specifically leukemia, lymphoma, or sarcoma; (b) diagnosis and treatment at Connecticut Children’s Medical Center (CCMC), Hartford Hospital, or Yale New Haven Hospital; and (c) age at diagnosis of 15–34 years. Patients who did not speak English or had a life expectancy estimated by their primary oncologist as being less than six months were excluded. The RCA is an agent of the Connecticut Tumor Registry designated by the commissioner of public health to facilitate cancer studies in Connecticut and has been used in previous studies (Yale Cancer Center, n.d.).

The study was reviewed and approved by the Connecticut Department of Public Health Human Investigation Committee, the Yale University Human Investigation Committee, and the institutional review boards (IRBs) of CCMC and Hartford Hospital.

Recruitment and Baseline Assessment
At CCMC and Hartford Hospital, after physician permission was granted to contact patients, a packet was mailed to the AYAs or to the parents of the AYAs aged younger than 18 years. The packet contained information describing the study and provided the IRB-approved adult consent form or parental permission and adolescent assent form, a medical record release form with Health Insurance Portability and Accountability Act authorization for medical record abstraction, and a self-addressed stamped envelope. Once the signed documents were received, surveys were sent. For nonrespondents to the baseline survey, a second mailing was sent.
(maximum of two attempts). Telephone calls were made (maximum of three attempts) to reach AYAs or parents. Participants completed the same mailed survey at 18 months postdiagnosis. A second mailing and as many as three telephone reminders were given for those who did not return a completed follow-up survey.

**Measures**
Baseline and follow-up surveys assessed demographics and health concerns. Diagnosis and treatment information was collected by the RCA, and medical record abstraction was conducted about 12 months postdiagnosis to collect data about diagnostic procedures, pathology reports, tumor characteristics, staging information, comorbid conditions, chemotherapy agents and cumulative doses, and radiotherapy (e.g., field, delivery mode, dose).

**PSYCHOLOGICAL DISTRESS**
The Brief Symptom Inventory (BSI), a 53-item self-report tool, was used to measure psychological distress (Derogatis & Spencer, 1993). This tool has demonstrated reliability and validity in samples comprised of AYA cancer survivors (Recklitis et al., 2006). Each item is rated on a five-point scale of distress ranging from 0 (not at all) to 4 (extremely). The BSI measures nine symptom dimensions of distress compiling three global indices. The current authors analyzed the three previously validated subscales in this population: depression, anxiety, and somatization (Recklitis et al., 2006; Zeltzer et al., 2009). The Global Severity Index (GSI), which is the summary score for global distress shown to be the best indicator of current distress and good reliability, was also calculated (Cronbach alpha of 0.9). An adjusted GSI score of 63 or greater implies clinically significant psychological distress relative to age and gender norms (Derogatis & Spencer, 1993).

**SURVIVORSHIP CARE AND THE SURVIVORSHIP CARE PLAN**
At both survey time points, AYAs indicated their preferences about where they wished to receive survivorship care (e.g., primary care physician’s office, specialty survivorship clinic). For those who had completed cancer therapy, at 18 months postdiagnosis, a personalized survivorship care plan (SCP) was given to the AYAs and their oncologists (or another physician specified by the AYA or his or her parent), with a survey evaluating its helpfulness, format, and content. The SCP consists of a treatment summary, recommendations for risk-based follow-up surveillance, and information about healthy lifestyle behaviors that can help control the risk for treatment late effects.

**Analysis**
The demographic and diagnostic characteristics of participants and nonparticipants were compared based on data collected by the RCA. Feasibility data (e.g., response rate, completion rate at each time point, attrition rate [and reasons] at each time point) were reported using descriptive statistics and compared to the range of response rates found in the literature. Frequencies of endorsing certain health concerns and experiencing clinically significant psychological distress at baseline and follow-up were calculated and compared to BSI normative data for community samples (GSI cut-point of 63). Paired t tests compared mean adjusted psychological distress scores at baseline and follow-up. A significance level of 0.05 (two-tailed) was used for all analyses. All analyses were conducted using SAS®, version 9.3.

**Findings**
A total of 129 AYAs identified through the RCA were assessed for eligibility. The majority of those identified were at Yale New Haven Hospital.
Hospital (n = 97), with 16 AYAs at Hartford Hospital and 16 at CCMC. Seven AYAs were determined to be deceased, and physician permission to contact was denied for three, leaving 119 eligible AYAs.

Demographics and Generalizability of the Sample
Participants were similar to nonparticipants in terms of race, age, and treating hospital. Of the 119 eligible AYAs, 94 did not enroll in the study; of these, 19 patients aged 18 years or older actively declined, 16 parents of patients aged younger than 18 years declined on their child’s behalf, and 59 did not respond to repeated messages. Overall, 25 patients enrolled in the study and completed the baseline survey (see Table 1). Participants were aged 16–34 years, with a mean age of 23 years (SD = 6). Twenty were aged 18 years or older, and 5 were aged younger than 18 years. In addition, 13 participants were male, and 12 were female. Twenty had private insurance, and of these, 15 were insured through an employer, either their employer (n = 8) or a parent’s employer (n = 7).

Feasibility of Recruitment and Retention
Of the 25 of 119 eligible AYAs who consented to participate, one consented in person at CCMC, and 24 participants or their parents returned signed consents by mail. Among the three sites, 17 of 96 eligible participants at Yale New Haven Hospital consented, compared to 4 of 16 at both Hartford Hospital and CCMC. Of the 25 participants who completed the baseline survey, three relapsed and one died, leaving 21 eligible for follow-up. Of these, three were lost to follow-up, despite repeated attempts to contact them. Eighteen completed the follow-up survey.

Concerns, Psychological Distress, and Care Preferences

HEALTH CONCERNS
At baseline, 24 participants were concerned about their future health, 20 about developing a subsequent or recurrent cancer, 19 about possible late effects of their treatments, 17 about their ability to have children, and 12 about accessing health insurance. At 18 months postdiagnosis, fewer participants reported concern about future health (n = 13, p = 0.54), developing another cancer (n = 13, p = 0.83), and accessing health insurance (n = 6, p = 0.53).

PSYCHOLOGICAL DISTRESS
At baseline, 10 participants experienced clinically significant somatization symptoms, and 9 experienced hostility, 8 experienced anxiety, 5 experienced depression, and 7 experienced global psychological distress (measured by the GSI), as indicated by BSI community norms for age and sex. No statistically or clinically significant changes were observed in mean standardized scores for any symptom subscale from baseline to follow-up (see Table 2).

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The SCP was sent to the 21 participants who had completed cancer therapy by the end of the study. Fourteen participants rated the helpfulness of the SCP. Of these, 10 noted that the SCP was very or somewhat helpful. Regarding the SCP’s format and distribution, 14 preferred a hard copy versus an electronic version (e.g., website, memory stick). In addition, seven indicated that the SCP alerted them to potential late effects of which they had been unaware.

After participants received the SCP, an increase was noted in the number of participants indicating they would prefer to receive survivorship care from an oncologist (from 15 of 25 to 12 of 18) or a survivorship clinic (3 to 5). In addition, a decline (from 8 to 4) was observed in those preferring to receive survivorship care from their primary care provider (PCP). SCPs and follow-up surveys were sent to physicians (16 oncologists and 3 PCPs) designated by the 19 participants who agreed to have their SCP shared. Only

### TABLE 2.
MEAN PSYCHOLOGICAL FUNCTIONING (BRIEF SYMPTOM INVENTORY) SCORES AT BASELINE AND AT FOLLOW-UP

<table>
<thead>
<tr>
<th>VARIABLE</th>
<th>BASELINE (N = 25)</th>
<th>FOLLOW-UP (N = 18)</th>
<th>p*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>X</td>
<td>SD</td>
<td>FD (%)</td>
</tr>
<tr>
<td>Anxiety</td>
<td>54.3</td>
<td>13.7</td>
<td>7</td>
</tr>
<tr>
<td>Depression</td>
<td>54.9</td>
<td>12.53</td>
<td>5</td>
</tr>
<tr>
<td>GSI</td>
<td>55.5</td>
<td>12</td>
<td>7</td>
</tr>
<tr>
<td>Somatization</td>
<td>61.1</td>
<td>12.5</td>
<td>10</td>
</tr>
</tbody>
</table>

*The p value was calculated by paired t tests to compare mean scores.
FD—frequency distressed; GSI—Global Severity Index (composite score).
Note. For FD, scores of 63 or greater were categorized to have clinically significant symptomology of distress (range of 30–80).
eight oncologists and one PCP responded to the survey, despite mail and email reminders. Most (seven oncologists and one PCP) found the SCP to be helpful. Four oncologists reported that they incorporated the SCP into the AYAs’ follow-up plan, and three oncologists discussed the SCP with the patient at his or her next clinical visit. Two oncologists kept the SCP on file. Four physicians preferred that patients receive follow-up care in the PCP’s office, whereas three oncologists and one PCP preferred that follow-up occur in the office of an oncologist, and one oncologist preferred that it take place in a survivorship care clinic.

Discussion

Substantial challenges were identified in recruiting AYA patients to this survivorship study. Although the current authors anticipated that early recruitment of AYAs (within six months of diagnosis) would be helpful, only 25 participants consented and completed the baseline survey, resulting in a suboptimal (21%) participation rate. Participation rates in epidemiologic studies have been declining (Ramos et al., 2015), and recruiting AYAs to survivorship-related studies presents specific challenges because of their unique psychosocial needs, developmental challenges, and mobility (Cantrell et al., 2012; Clinton-McHarg et al., 2011; Kazak et al., 2010).

This study implemented an innovative strategy in terms of starting patient recruitment within six months of diagnosis; previous studies have recruited AYAs one or more years post-diagnosis (Gorman et al., 2014; Kazak et al., 2010). However, early recruitment failed to result in participation rates higher than those previously reported by Harlan et al. (2011) (43%), Hendricks-Ferguson et al. (2013) (50%), Link et al. (2009) (41%), and Richards et al. (2010) (61%), suggesting it is not a feasible strategy. Physician permission did not present a barrier in the current study, with 99% of physicians permitting contact with eligible patients. This contrasts with low rates of physician permission reported in Clinton-McHarg et al. (2011) (reduction in eligibility from 411 to 232).

Although the current authors expected the RCA system to improve responses by limiting missing or incorrect contact information, a considerable proportion of those identified could not be contacted. Given the challenges involved in locating patients within six months of diagnosis, AYA survivorship may require strategies such as in-person recruitment and effective tracing procedures. Although the contact information available was likely up to date, because the AYAs were being treated at the study sites, many neglected to respond to mail or telephone contact. With the shift from paper to electronic mail and the option to screen telephone calls, particularly those from unknown callers during a challenging time in the cancer trajectory, recruitment of AYAs may require even more innovative strategies.

As of 2011, 95% of AYAs aged 18–29 years reported using the Internet, and 72% reported searching online for health information. Sixty-two percent of smartphone users have used their phone in the past year to look up information about a health condition (Smith, 2015). In addition, 52% of smartphone users gather health information via their smartphone (Fox, 2013; Siminoff, 2008). Internet recruitment has previously been identified as cost effective and successful (Rabin et al., 2013), and social media has been identified as a potentially reliable recruitment method (Gorman et al., 2014). Future studies may evaluate the effectiveness of these AYA-friendly strategies to maintain up-to-date contact information for a geographically mobile population and increase AYAs’ participation and response rates in survivorship research.

The sample size may have been limited by not directly involving the AYAs’ physicians or oncology nurses in recruitment. Although all physicians were contacted for permission to approach their patients, oncology providers were not responsible for introducing the study to the AYAs. Previous studies have shown that direct contact from the attending physician can assist with recruitment (Siminoff, 2008). In addition, given the extent and nature of their contact with patients, oncology nurses and nurse practitioners are untapped resources for facilitating recruitment and retention of AYAs in survivorship research. Researchers, including those who are oncology nurses, may improve recruitment of AYAs to survivorship research by engaging nurses and nurse practitioners in study design and implementation.

In terms of psychological distress, the results indicate high frequencies of psychological distress, including somatization, anxiety, depression, and hostility among the participants at baseline. No statistically significant differences were noted, including no improvement in mean scores from baseline to follow-up, which could suggest persistent psychological distress or be attributable to the small sample size. Future research is needed to determine if these results hold up in a larger sample, because persistent psychological distress 18 months postdiagnosis would indicate the need for a full assessment by mental health professionals and treatment as indicated. Recent definitions of AYAs extend upward to age 39 years (NCI, 2015); consequently, some patients who otherwise would have been included in a study of AYAs were excluded, which also limited the sample size.

The sample in this study showed decreased health concerns from baseline to follow-up; however, this change was not
statistically significant, and the sample was small. Previous studies in older adults have shown the contrary, with long-term survivors experiencing persistent health concerns (Warner et al., 2016) in addition to worries about health insurance and finances (Zebrack, Kent, Keegan, Kato, & Smith, 2013). This discrepancy could be a result of the small sample size, indicate perceptions of invincibility among AYAs, or stem from oncology social worker assistance with accessing health insurance and financial support. Despite treatment exposures and risk for late effects, most AYAs had fewer worries about future health, late effects, second cancers, and ability to have children at follow-up than at baseline. This notion of perceived invincibility has previously been acknowledged (NCI, 2006) and could be a strategy for managing distress and uncertainty about the future by limiting cognitive awareness of future health problems. A larger, more representative sample is needed to determine if higher concerns at baseline were a result of participation bias—that is, those most concerned about their health tended to enroll. Alternatively, concerns at diagnosis could be even greater in the general population of AYAs diagnosed with cancer, because worries about health have been shown to deter participation in health-related research (Helmes, Bowen, Bowden, & Bengel, 2000). Because the sample size of this study was not sufficiently powered to measure change, the results are hypothesis generating for future research, but cannot be directly interpreted.

Results of the follow-up survey revealed that five participants would prefer to receive survivorship information by email, whereas nine would prefer to receive hard-copy information by mail. Despite finding the survivorship materials very or somewhat helpful, seven participants replied that they “did nothing with them,” and two “put them away for later.” These are behaviors that, as with their reports of fewer worries about health, suggest attempts to limit cognitive awareness of distressing information, such as potential late effects. The motivations for these behaviors could be explored in future research. Such research could be a step toward developing interventions that address these behaviors in an attempt to provide quality care for survivors, including information about potential late effects and enactment of the recommendations. Despite their decreasing health concerns, AYAs, as well as their physicians, noted that the SCP was helpful. AYAs stated that SCPs should be shared between oncology providers and PCPs, which suggests some element of concern about future health. Using the SCP to disseminate information about risk-based follow-up care should continue to be investigated for impact on implementation of the recommendations stated in the SCP and effectiveness of patient education on the value of survivorship care.

Implications for Nursing
As members of the multidisciplinary team, oncology nurses and nurse practitioners must be aware of the challenges inherent in advancing research and evidence-based care of AYAs who have been diagnosed with cancer. Given their extensive personal contact with patients, oncology nurses and nurse practitioners must develop sensitivity to psychosocial distress and health concerns among patients in the AYA age range, initiate referrals for further assessment of those exhibiting distress, and solicit and address their health concerns. When AYAs exhibit concerns about healthcare costs and access to insurance, recommendations are for oncology nurses and nurse practitioners to guide them to available resources and collaborate with social workers to help AYAs navigate access. In addition, oncology nurses and nurse practitioners should be involved in preparing and distributing SCPs, providing education about potential treatment side effects and longer-term complications of AYA cancer, and communicating with AYAs about the importance of adopting healthy lifestyle behaviors, including regular engagement in health care. The work of preparing treatment summaries and the other elements of the SCP has become less labor- and time-intensive with the advent of electronic health records that include oncology-specific elements. To advance survivorship care and research, oncology nurses should maintain up-to-date contact information for their AYA patients, inform their patients meeting eligibility criteria about opportunities for participation in research, link interested parties with the research team, and guide these AYAs in developing questions to ask during the informed consent process. In addition, oncology nurses and nurse practitioners should actively engage in AYA survivorship research as members or leaders of studies that address barriers to participation (psychological distress and other symptoms) and use the social media platforms and digital technologies that are part of AYAs’ lives to promote their recruitment and fulfillment of study requirements.

Conclusion
From this pilot study, the current authors determined that early recruitment may not be a means to improve AYAs’ participation in survivorship research. Unique strategies are needed to recruit, enroll, and retain this highly mobile, hard-to-reach population of patients with cancer. A large proportion of newly diagnosed AYAs experiencing health concerns and psychological distress were identified. In addition, many survivors and their physicians are receptive to the SCP. As the field of AYA oncology continues to develop, new studies with innovative methods for recruitment are necessary to enhance AYAs’ access to survivorship care.
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