Promoting Resilience in Stress Management: A Pilot Study of a Novel Resilience-Promoting Intervention for Adolescents and Young Adults With Serious Illness

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Abstract

Objective To examine the feasibility and format of the Promoting Resilience in Stress Management (PRISM) intervention among two groups of adolescents and young adults (AYAs) at-risk for poor outcomes: those with Type 1 diabetes (T1D) or cancer. Methods PRISM consists of two long or four short skills-based modules. English-speaking patients 12–25 years old were eligible if they had T1D for >6 months or cancer for >2 weeks. Feasibility was defined as an 80% completion rate and high satisfaction. Ongoing monitoring shaped iterative refinement of disease-specific approach. Results 12 of 15 patients with T1D (80%) completed the two-session intervention. 3 of 15 patients with cancer declined to complete the two-session version, citing prohibitive length of individual sessions. 12 (80%) completed the four-session version. Patient-reported satisfaction was high across groups. Conclusions The PRISM intervention is feasible and well-accepted by AYAs with cancer or T1D. Differences in patient populations warrant differences in approach.

Key words: adolescent young adult; AYA; cancer; coping; diabetes; health outcomes; intervention; psychosocial outcomes; resilience.

Introduction

The experience of serious illness among adolescents and young adults (AYAs) is unique. Youth in this age-group must learn to cope not only with the demands of their illness but also with normal developmental challenges, transitions, and choices related to independence, education, employment, relationships, family, and personal identity (Eiser, Penn, Katz, & Barr, 2009). Two distinctive examples of populations that may struggle with these collective challenges are AYAs with cancer or Type 1 diabetes (T1D). Both groups report significantly elevated stress: Cancer survivors who were diagnosed as AYAs have greater psychological distress and fewer positive health beliefs than younger pediatric or older adult survivors (Kazak et al., 2004, 2010). More than 50% do not pursue adequate cancer-related follow-up care (Nathan et al., 2008), and ongoing physical, social, and emotional challenges such as physical impairment, infertility, uncertainty, fears about recurrence, interruption of life
plans, and discrimination in employment and insurance are common (Pendley, Dahlquist, & Dreyer, 1997; Richardson, Nelson, & Meeske, 1999). Similarly, the prevalence of depression is two to three times greater in AYAs with T1D than in nondiabetic youth (Grey, Whittemore, & Tamborlane, 2002; Kovacs, Goldston, Obrosky, & Bonar, 1997). AYAs with T1D struggle with poor adherence to treatment plans (Borus & Laffel, 2010) and high levels of diabetes-related distress (Weissberg-Benchell & Antisdel-Lomaglio, 2011), leading to high rates of preventable complications such as diabetic ketoacidosis (Shrestha, Zhang, Barker, & Imperatore, 2010). Glycemic control is notoriously poor during this period; only 30% of adolescents with T1D meet their glycemic targets (Petitti et al., 2009).

Although specific illness-related challenges may differ between these two groups of patients, potential strategies to overcome them may transcend diagnosis. Resilience, for example, is a universal construct describing an individual’s capacity to maintain psychological and/or physical well-being in the face of stress, and is a good candidate to buffer the negative impact of serious illness among multiple populations of AYAs (Haase, 2004; Southwick & Charney, 2012). Unfortunately, variable theories and factors of resilience have precluded systematic approaches to intervention development (Molina et al., 2014). Few studies have described AYA-specific perspectives of resilience (Gillham et al., 2007; Haase, 2004); to our knowledge, there are no age-appropriate interventions designed to promote it among AYAs with serious illness.

The objective of this study was to develop and refine a resilience-building intervention for AYAs with serious illness through iterative pilot testing of feasibility and patient-reported acceptability. We specifically aimed to create an intervention that would span multiple types of illness experience. We also anticipated that successful implementation of an intervention might vary by patient population. Hence, we conducted parallel pilot studies among two groups of AYAs at high risk of elevated stress and subsequent poor long-term outcomes: those with T1D or those with cancer. Exploratory aims were to collect additional pilot data regarding patient-reported resilience such that future larger studies could be appropriately designed. Ultimately, our goal is to offer a practical intervention to bolster resilience in the context of chronic disease management, thereby improving long-term psychosocial outcomes.

Methods

Intervention Development

The Promoting Resilience in Stress Management (PRISM) intervention was developed through three primary stages. First, we conducted a comprehensive review of published theoretical models of resilience among AYAs with cancer (Haase, 2004; Ishibashi et al., 2010; Robb et al., 2014) and other resilience-to-stress models (Burton, Pakenham, & Brown, 2010; Folkman & Greer, 2000; Southwick & Charney, 2012). In addition, we identified and examined positive psychological and/resilience-based interventions that exist for well AYAs and adult populations with serious illness. Evidence suggested four specific, teachable skills for intervention development: (1) stress management, where interventions were associated with reduced depression (Carver, 2005; Gillham et al., 2007; Van Son et al., 2012); (2) goal-setting and problem-solving, which were associated with psychosocial well-being (Carver, 2005; McCarty, Violette, & McCauley, 2011); (3) positive re-appraisals of stressors, which were shown to reduce distress and improve adherence and quality of life (Bradshaw et al., 2007; Gillham et al., 2007; McCarty et al., 2011; Steinhardt, Mamerow, Brown, & Jolly, 2009; van der Lee & Garssen, 2012); and (4) meaning-making or benefit-finding, which were associated with improved quality of life (Casellas-Grau, Font, & Vives, 2013; Henry et al., 2010).

Second, we solicited expert and stakeholder opinion by presenting potential interventions-in-development at national meetings (Rosenberg et al., 2013a, 2013b) and interviewing AYA patients, physicians, social workers, psychologists, nurses, and other psychosocial clinicians involved in the care of seriously ill AYAs. Consensus was that a successful intervention would teach resilience-building skills; however, the intervention would have to be brief, inexpensive, and easily adaptable to ensure feasibility.

Third, we considered findings from our prior and ongoing studies of resilience among AYAs with cancer and T1D. For example, AYAs with cancer consistently reported needing or developing the same four key skills mentioned above to adjust to, cope with, and ultimately recover from their illness experience (Rosenberg, Yi-Frazier, Wharton, Gordon, & Jones, 2014). Among patients with diabetes, low perceptions of personal resilience resources were associated with high distress, poor quality of life, and poor glycemic control (Yi-Frazier et al., 2010, 2015).

Intervention Design and Delivery

These steps led to the first authors to create and manu- alize the PRISM intervention, which targeted the four above-described disease-non-specific skills (Table I). It was originally designed as two main 30–50-min sessions, administered 2–4 weeks apart. These were followed by a follow-up session (approximately 30 min) to reflect on the skills learned and resources needed. This brief format was selected based on stakeholder
feedback and literature suggesting AYAs preferred brief interventions to longer, traditional cognitive behavioral therapy (CBT) interventions. (Steinhardt & Dolbier, 2008) To further aid skills development, participants were given “cheat sheets” or handouts describing each skill and how to practice the techniques. No specific homework was assigned. The intervention was administered directly to the patient; however, parents were invited to participate by listening to the two primary sessions at the patients’ discretion.

The first session was delivered in-person, often in conjunction with a clinic visit (patients with cancer or diabetes) or in the inpatient setting (patients with cancer). Any subsequent sessions could be administered in-person, by Skype, or by phone. One interventionist was assigned to each family based on availability; only this person was present with each family during each session. To facilitate future dissemination, three clinical research associates (bachelor’s or master’s level) were trained to administer the intervention. Each completed 8–12 hr of training including supervised role-playing and practice scenarios with the principal investigators and social workers from the diabetes and oncology teams. Each administration was audio-recorded and subsequently evaluated for fidelity using a scoring instrument developed for the present study.

Pilot Feasibility and Acceptability Study
To optimize future implementation of the intervention, we examined patient-preferred and disease-specific approaches with an iterative pilot feasibility and acceptability study.

Participants
Consecutive English-speaking patients were approached in the diabetes clinic and the cancer clinic or in-patient ward, respectively. They were eligible if they were between 12 and 25 years old and received their cancer or diabetes care at Seattle Children’s Hospital (SCH). Patients with cancer had been diagnosed for at least 2 weeks, and those with T1D for at least 6 months. Patients with cancer were included earlier after their diagnosis based on explicit patient-requests during formative interviews. For patients with diabetes, those newly diagnosed were not included, as they face significantly different stressors than established patients, as they are still adjusting to their daily self-care management. All patients were offered a $35 gift card as compensation for their time and feedback. Patients ≥18 years old provided signed informed consent; those <18 years provided signed assent and their parents provided signed consent. All patients were asked whether they wanted their parents (or other primary caregivers) to participate; if so, then parents provided additional consent for their own participation. The study was approved by the SCH institutional review board.

Demographic Variables
Basic self-reported demographic variables (sex, age, cancer diagnosis) were collected from all participants at the time of enrollment. These were summarized descriptively and comparisons between diabetes and cancer participants were conducted using independent samples t tests or chi-squared tests.

Outcomes
We defined feasibility as 80% completion of all main sessions among enrolled participants. The iterative design incorporated ongoing real-time assessments of enrollment and completion rates, as well as an option to revise the intervention format based on mid-study feasibility and acceptability results. We defined acceptability based on qualitative patient and parent feedback following each session and at the follow-up session. Interviews were audio-recorded, transcribed verbatim, and directed at the intervention content, timing, duration, and delivery. In addition, we queried participants regarding unmet needs (i.e., the need for a separate parent-directed intervention), and willingness
to participate in additional studies (i.e., in further development of family adaptations of the PRISM).

**Exploratory Analyses**

This pilot study was not designed or powered to test intervention efficacy; however, to collect data for future clinical trials, each participant was invited to complete a brief survey at the time of enrollment and at the time of the follow-up session. Patient-reported resilience was measured with the 10-item version of the Connor–Davidson Resilience Scale (CD-RISC; Campbell-Sills & Stein, 2007; Connor & Davidson, 2003). This instrument has excellent psychometric properties (Cronbach’s $\alpha = .85$) and has been validated in AYA populations (Campbell-Sills, Cohan, & Stein, 2006), as well as patients with cancer (Scali et al., 2012) or diabetes (Steinhardt et al., 2009). Higher scores suggest higher personal resilience resources, and prior studies have shown that lower scores are associated with inferior psychosocial outcomes (Rosenberg, Wolfe, et al., 2014), and that interventions may increase individual scores (Davidson et al., 2006). Among patients for whom both baseline and follow-up scores were available, change scores were calculated by subtracting baseline scores from follow-up scores; differences between disease groups were explored through independent $t$ tests. Paired $t$ tests were conducted to detect changes from baseline to follow-up on the CD-RISC total scores. Effect sizes were calculated for both disease groups individually and combined.

**Results**

**Group-Wide Results**

Feasibility varied by diagnosis; however, qualitative comments regarding acceptability were similar across groups. Common reasons for not enrolling were being “too busy” or “not interested.” There were no differences between groups on any of the baseline variables (Table II). Feedback from participants was universally positive. AYA participants made comments such as: “This is so helpful, I wish we had done this sooner,” “I was actually telling [my friends] about it and they said they would try it out,” and “I think [they’re] good techniques to use, definitely. I’m teaching my little sister. I’m sure it can help her too.” Parents in both groups endorsed high satisfaction with the intervention, optimism that it would help their children, and requests for resources for continued skills-training. Both patients and parents in each group suggested that a separate parent-focused version of the intervention would be helpful.

**AYAs With T1D**

Fifteen of 26 patients with diabetes (58%) agreed to participate and signed informed consent (Table II). All first sessions were delivered in-person; 77% of following sessions were conducted by telephone, based on patient requests to complete the intervention without having to come to the hospital for a nonmedically indicated visit. Thirteen family members agreed to participate in the first session; however, only five participated in the second. Of the 15 enrolled patients, 1 was lost to follow-up, and 12 (86% of those remaining, 80% of those enrolled, 46% of those approached) completed all core PRISM sessions. The median number of days between sessions was 11 (interquartile range [IQR] = 11–20). No patient offered additional suggestions regarding how to improve enrollment, and feedback was highly positive, including satisfaction with the format. Participants who completed the intervention stated they would prefer the sessions delivered as received. Thus, no changes were made to the diabetes-specific approach.

**AYAs With Cancer**

Fifteen of 22 patients with cancer (68%) enrolled, including 12 with their family members (Table II). However, initial retention was challenging. Three

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Diabetes</th>
<th>Cancer</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>15.1 ± 1.3</td>
<td>16.2 ± 2.8</td>
</tr>
<tr>
<td>Sex (% female)</td>
<td>67</td>
<td>58</td>
</tr>
<tr>
<td>Race (% White)</td>
<td>67</td>
<td>75</td>
</tr>
<tr>
<td>Ethnicity (% non-Hispanic White)</td>
<td>80</td>
<td>75</td>
</tr>
<tr>
<td>Duration of disease (years)</td>
<td>5.2 ± 4.7</td>
<td>2.3 ± 3.8</td>
</tr>
<tr>
<td>CD-RISC (mean)</td>
<td>30.1 ± 4.2</td>
<td>27.3 ± 3.9*</td>
</tr>
</tbody>
</table>
| Type of cancer: n (%)   | N/A      | Sarcoma (initial diagnosis): 5 (33)
|                         |          | Acute leukemia (initial diagnosis): 3 (20)
|                         |          | Acute leukemia (first recurrence): 3 (20)
|                         |          | Sarcoma (first recurrence): 2 (13)
|                         |          | Hodgkin lymphoma: 1 (7)
|                         |          | Germ cell tumor: 1 (7) |

*Note. CD-RISC = Connor–Davidson Resilience Scale.
* $n = 12.$
patients dropped out early on, stating that it was difficult to dedicate the proper amount of uninterrupted time to the hour-long sessions, due to conflicting medical procedures or illness symptoms. In addition, after the first session, many AYAs with cancer requested that family members not participate in future sessions, stating parents were a distraction. Qualitative explanations were that it was more effective to talk about stresses and worries without the added worry of burdening parents. Only one family member of a patient with cancer returned for the second session; in all other cases, the AYA, the parent, or both suggested the sessions would be more productive without concurrent parent participation. The median number of days between sessions was 18 (IQR = 17–67).

For these reasons, the format of the intervention for patients with cancer was revised from two 50-min sessions with optional parent participation to four 30-min, one-on-one sessions that excluded parents. Subsequently, of the 12 remaining patients, all (100% of those remaining, 80% of those enrolled, and 54% of those approached) completed the intervention. The median number of days between sessions was similar (median = 21, IQR = 11–64). All sessions for cancer were delivered in-person, in conjunction either with inpatient hospitalizations or outpatient clinic visits, based on patient preferences for convenience.

### Interventionists

Three interventionists, all nonmedical research staff with college degrees (one with a master’s in counseling), successfully delivered the PRISM intervention at least once. Fidelity scores were 100% for delivering the appropriate content. Qualitative feedback for the ability to deliver the intervention as outlined via the protocol was positive, there were no differences in feedback based on intervenor, and no abnormal psychosocial or emotional issues arose such that consult was needed from a social worker or psychiatrist.

### Exploratory Analyses

Follow-up data on resilience were available from 21 AYAs participants (n = 12 diabetes, n = 9 cancer; Table III). Pooled and stratified mean CD-RISC scores did not change. Effect sizes indicated a potential, small effect, particularly for the cancer group. No differences were found between disease group in change scores.

### Discussion

To our knowledge, this is the first description of a resilience-promoting intervention for AYAs that spans multiple types of serious illness. We found that the intervention was highly valued by both patients and their parents. Qualitative comments suggest the PRISM or similar interventions designed to support coping and adjustment may be critical components of whole-patient care and echo those of national organizations like the Institute of Medicine (IOM, 2007). Patients and families in our study universally confirmed that stress management, goal-setting, positive reframing, and benefit-finding were all skills that could (and should) be taught to patients with serious illness.

Feasibility of the intervention varied by patient diagnosis. We found that individual patient populations required specific adaptations of the intervention and that flexibility of the curriculum enhanced participation. Among AYAs with T1D, initial enrollment was challenging and fell short of our anticipated accrual. Still, among the patients who did enroll, 80% completed the two-session version of the intervention and endorsed it for future patients. Patients with T1D were generally able to complete the sessions within the 2–4-week window as proposed. Among AYAs with cancer, enrollment rates were sufficient, but high rates of early dropout and widely variable periods between sessions prompted changes in approach to allow shorter and more frequent sessions. Subsequent to those revisions, both enrollment and retention remained excellent; however, time between sessions remained unchanged. Future studies of the intervention among AYAs with cancer will incorporate additional procedures to standardize time between sessions. In both groups of patients, we found that patient preferences and convenience defined success.

This study was not designed to detect intervention efficacy; we conducted exploratory analyses to gather preliminary data for future studies. As such, we did not identify statistically significant changes in patient-reported resilience and only noted small to intermediate effect sizes across groups. The most likely explanation for these results is that the study was

<table>
<thead>
<tr>
<th>Study group</th>
<th>Baseline CD-RISC (SD)</th>
<th>Follow-up CD-RISC (SD)</th>
<th>Change in mean scores</th>
<th>p-value</th>
<th>Effect size</th>
</tr>
</thead>
<tbody>
<tr>
<td>Combined</td>
<td>27.9 (3.8)</td>
<td>29.8 (6.0)</td>
<td>1.9 (4.9)</td>
<td>.097</td>
<td>.23</td>
</tr>
<tr>
<td>T1D (n = 12)</td>
<td>29.2 (3.5)</td>
<td>30.8 (6.0)</td>
<td>1.6 (5.2)</td>
<td>.315</td>
<td>.15</td>
</tr>
<tr>
<td>Cancer (n = 9)</td>
<td>26.2 (3.7)</td>
<td>28.4 (6.0)</td>
<td>2.2 (4.7)</td>
<td>.195</td>
<td>.32</td>
</tr>
</tbody>
</table>
et al., 2014). While stress and coping theories confirm (Folkman & Greer, 2000), a practical challenge has that the PRISM skills are amenable to interventions. These are particularly challenging for AYAs with serious illness. The average refusal rate for traditional CBT interventions in pediatric chronic disease settings is 37%, with attrition rates up to 32% (Karlson & Rapoff, 2009). Notably, we found similar rates of refusal with our skills-based format, suggesting even abbreviated interventions are challenging. In contrast, others have found that shorter skills-based interventions may be more successful (Steinhardt & Dolbier, 2008).

An additional consideration for AYA interventions is the fact that parent and family interactions contribute to patient outcomes. For example, patient and/or parent distress can hinder communication with the medical team, therefore affecting decision making (Burns, Robb, Phillips-Salimi, & Haase, 2010). Moreover, joint advanced care planning interventions that target AYAs but also include parents have been shown to promote communication and realistic goal-setting (Lyon et al., 2009). Our study attempted to include parent involvement from the beginning; however, we found that AYAs with cancer tended to prefer conducting the intervention without their parents, whereas youth with T1D seemed to appreciate parent presence. Nevertheless, parent involvement in the second T1D session may have been minimal because many of these patients participated by phone. In both cases, parents endorsed a wish for their own “parent-adapted” version of the PRISM intervention.

Targeted interventions with AYAs with cancer (Burns et al., 2010; Haase, 2004) and T1D (Laffel et al., 2003; Nansel et al., 2009) show promise, as do those aimed at general parenting (Perrin, Sheldrick, McMenamy, Henson, & Carter, 2014) or specific parent problem-solving skills (Sahler et al., 2013). We aimed to design an intervention that could be adapted for several pediatric settings and demonstrated that our brief, skills-based format was not only feasible but also highly valued by patients and families. Ongoing studies include the development of parent-directed PRISM modules as well as larger studies exploring the efficacy of the PRISM on patient outcomes in AYAs with cancer and T1D. Future studies may include adaptations for other serious pediatric conditions, such as cystic fibrosis, chronic kidney disease, rheumatologic disease, obesity, and traumatic brain or spinal cord injuries. Ultimately, this research may lead to improved patient (and family) long-term wellness after the life-changing diagnosis of serious condition.

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