

Randomized Clinical Trial of a Legacy Intervention for Quality of Life in Children with Advanced Cancer

Terrah Foster Akard, PhD, RN, CPNP, FAAN,¹ Mary S. Dietrich, PhD, MS,²
Debra L. Friedman, MD, MS,³ Sarah Wray, MSN, RN, CPNP,⁴ Cynthia A. Gerhardt, PhD,⁵
Verna Hendricks-Ferguson, PhD, RN, FPCN, FAAN,⁶ Pamela S. Hinds, PhD, RN, FAAN,⁷
Bethany Rhoten, PhD, RN,⁸ and Mary Jo Gilmer, PhD, MBA, RN-BC, FAAN⁹

Abstract

Background: Legacy-making (actions/behaviors aimed at being remembered) may be a significant component for quality of life (QOL) during advanced illness and end of life. Although legacy interventions have been tested in adults, the impact of legacy activities on QOL for children has yet to be clearly defined.

Objective: This study examined the impact of our newly developed web-based legacy intervention on dimensions of QOL among children (7–17 years old) with advanced cancer.

Design: This single-site randomized clinical trial (RCT) used a two-group waitlist control design. The legacy intervention guided children to create digital storyboards by directing them to answer legacy questions about themselves (personal characteristics, things they like to do, and connectedness with others) and upload photographs, video, and music.

Setting/Subjects: Facebook advertisements recruited children (ages 7–17) with relapsed/refractory cancer and their parents from the United States. Child-parent dyads ($N=150$) were randomized to the intervention or usual care group, and 97 dyads were included for analysis.

Measurements: Children and parents completed the PedsQL Cancer Module preintervention (T1) and post-intervention (T2).

Results: Although not statistically significant, legacy-making demonstrated small effects in child procedural anxiety and perceived physical appearance (Cohen's d 0.35–0.28) compared to the wait-list control group.

Conclusions: This study contributes important discoveries, including support for the feasibility of a RCT web-based legacy intervention for children with advanced cancer. We did not find convincing evidence supporting the hypothesis that legacy-making improved child dimensions of QOL across time. Overall, this is a null study that warrants discussion on possible reasons for limited findings. Future legacy intervention research is needed using qualitative and quantitative methods, as well as child and parent reports, to determine how such services may improve dimensions of QOL for pediatric palliative care populations. ClinicalTrials.gov number NCT04059393.

Keywords: Internet-based intervention; pediatric cancer; pediatric palliative care; quality of life

¹Vanderbilt University School of Nursing and School of Medicine, Nashville, Tennessee, USA.

²Vanderbilt University Schools of Medicine (Biostatistics, VICC, Psychiatry) and Nursing, Nashville, Tennessee, USA.

³Division of Hematology-Oncology, Vanderbilt University Medical Center and Vanderbilt-Ingram Cancer Center, Nashville, Tennessee, USA.

⁴Vanderbilt University School of Nursing, Nashville, Tennessee, USA.

⁵Department of Pediatrics and Psychology, The Ohio State University and Center for Biobehavioral Health, The Research Institute at Nationwide Children's Hospital, Columbus, Ohio, USA.

⁶Saint Louis University, School of Nursing, St. Louis, Missouri, USA.

⁷Department of Nursing Science, The William and Joanne Conway Chair in Nursing Research, Department of Pediatrics, Children's National Health System and The George Washington University, Washington, DC, USA.

⁸Department of Nursing, Vanderbilt University School of Nursing, Nashville, Tennessee, USA.

⁹Nursing and Medicine (Pediatrics), Vanderbilt University School of Nursing and Medicine, Nashville, Tennessee, USA.

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Introduction

CHILDREN WITH CANCER constitute a large number of children receiving pediatric palliative care. More than 15,000 children under 19 years of age are diagnosed with cancer in the United States each year, resulting in ~40,000 children receiving cancer treatment.¹ Children with cancer experience substantial suffering and decreased quality of life (QOL) throughout the illness trajectory, including physical symptom, emotional, and social distress.²⁻⁶ Symptom distress has been strongly associated with QOL in children with advanced cancer, with reported “poor/fair” categories in physical, school, emotional, and social subscales.⁷ The large majority of parents of children with advanced cancer have reported that they hope for their child’s QOL (e.g., happy, healthy, and enjoy life) and minimal suffering (e.g., least pain).⁸

Legacy-making, defined as actions or behaviors aimed at being remembered,⁹ may be a significant component for QOL during illness and end of life.¹⁰⁻¹³ Legacy activities include memory books, songwriting, photographs, and videos.¹²⁻¹⁴ Legacy-making has been explored in adult^{10,11} and pediatric populations^{9,12,13,15} and shown to increase patients’ sense of dignity, purpose, meaning, and will to live, while decreasing suffering and depressive symptoms.¹⁰ Hospital staff have reported that legacy activities helped ill children cope and communicate and their family members cope, communicate, and continue bonds in the case of the child’s death.¹² Legacy-making is a palliative care intervention that can also co-exist with curative care.^{9,12} Many hospitals currently offer legacy activities for children with serious illnesses¹²; however, the impact of legacy activities on ill children’s QOL has yet to be clearly defined. Children generally begin to understand that death is permanent and universal during Piaget’s concrete operational stage, which usually begins at seven years of age.¹⁶ Thus, school-age children are ideal to begin legacy discussions, consistent with evidence from our previous work in which most bereaved family members perceived children (ages 7–17 years) living with advanced cancer were aware of impending death.¹³

We previously developed a digital storytelling intervention that helped guide children with cancer to document

their legacies.^{9,15} Results indicated the intervention was feasible and had the potential to improve child QOL, but the face-to-face nature of the intervention limited our ability to access adequate sample sizes for rigorous intervention testing. Thus, we developed a web-based version of the legacy intervention to increase access and ultimately decrease suffering and enhance life for children with relapsed/refractory cancer and their parents. This article presents results that examine the impact of the web-based legacy intervention on QOL among children with relapsed or refractory cancer. We hypothesized that compared to usual care, legacy-making would improve dimensions of QOL among children across time.

Methods

Conceptual approach

We developed a conceptual framework (Fig. 1) based on synthesis of components of existing theories (e.g., Continuing Bonds Theory, Model of Dignity Therapy) and evidence^{10,17} to guide the proposed study.¹⁸ Briefly, this framework provided a basis for examining the direct effects of a legacy-making intervention on QOL for children living with refractory or relapsed cancer and their parents. This article includes results related to the study, primary outcome of child dimensions of QOL. Additional study outcomes (parent-child communication [primary outcome] and child and parent coping [secondary outcome]) will be reported in future articles.

Procedures

This research reports on one of the outcomes studied in a randomized clinical trial (RCT) examining effects of a legacy intervention on child and parent outcomes. Required institutional review board approval was obtained before subject enrollment. We aimed to enroll 150 child-parent dyads to achieve an analysis sample of $N=100$ to provide 80% statistical power (two-sided $\alpha=0.05$) for the detection of an intervention effect on the trajectories of child QOL as small as 0.30.

Advertisements were placed on Facebook over 3 years, targeting individuals who (1) were 13 years of age or older,

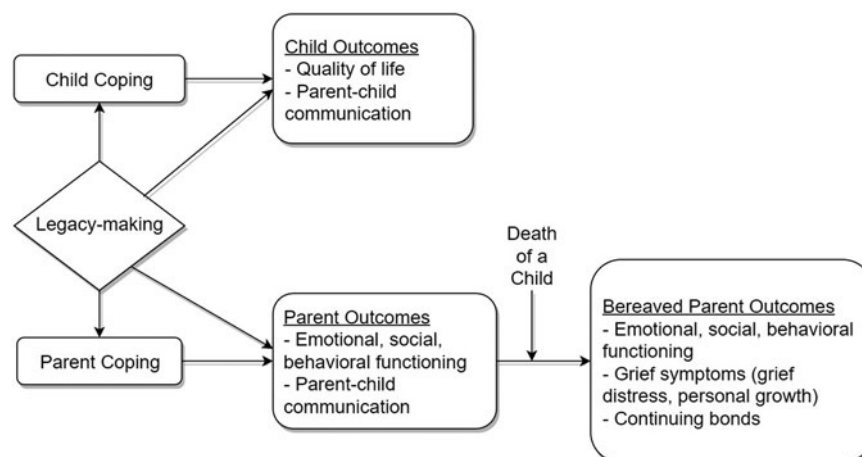


FIG. 1. Conceptual framework.

(2) were located in the United States, (3) were any gender, and (4) had interest in pediatric oncology (interest terms selected such as childhood cancer, and cure childhood cancer). Because Facebook users must be at least 13 years of age to have an account, our Facebook advertisements targeted individuals 13 years of age and older. This allowed us to access children (13 years of age or older) and parents through Facebook ads, with children younger than 13 years being accessed by their parents. Parents could click on an electronic REDCap link in the Facebook ad that briefly described the study and included initial screening questions.

Potentially eligible parents were then asked to complete some basic demographic questions, including their name and contact information, to receive more details about the study. The study coordinator contacted interested individuals through phone or e-mail within one week to describe the study and confirm eligibility. Eligible participants were (1) 7 to 17 years of age and their primary parent caregiver (i.e., legal parent guardian who spent the most number of hours per week with the eligible child), (2) patients with advanced cancer, defined as relapsed or refractory cancer determined by parent self-report, (3) able to speak, understand, read, and type English, (4) those with Internet access, and (5) those without cognitive impairment based on coordinator determination for parents during the consent process that they were cognitively able to understand and participate, and based on parent report for the child. Children 7 to 17 years of age were chosen because of their developmental understanding that death is permanent and universal and their likely awareness of impending death. The age of 17 years was selected as the cutoff because 18 years of age and older begin different developmental stages of independence.

For eligible participants, the coordinator obtained verbal parent consent and child assent and confirmed contact information to send study documents. Documentation of written consent and assent was waived as per the IRB. Upon consent, child and parent dyads were randomly assigned on a 1:1 ratio to either the intervention ($n=75$) or wait-list control ($n=75$) group by using a computer-generated randomization approach with a permuted block scheme generated by a PhD-prepared biostatistician. Neither participants nor study team members were blinded to participant assignment. The coordinator e-mailed an electronic link for the intervention website to participating parents in the *intervention group* within one week after children and parents completed baseline (T1) measures. Each child-parent dyad created a username and password to the website.

Legacy intervention

The intervention website guided children to create a digital storyboard about themselves by directing them to (1) *answer legacy questions about themselves*, including personal characteristics (e.g., name and age), things they like to do (e.g., hobbies and interests), and connectedness with others (e.g., special message to a loved one), (2) *upload photographs*, (3) *upload video*, and (4) *upload music*. Children could skip any question or content that they did not want to answer or include. They could complete the activity alone or request assistance from a family member. Children were asked to complete the intervention within two weeks. When children completed the storyboard, the website generated a unique

electronic link for the project coordinator to review the child's completed digital story. Once the coordinator approved the digital story as appropriate (e.g., no foul language or inappropriate photographs), she e-mailed the electronic link to the child or parent. Only individuals with the link were able to view the child's digital story. Part of the assent process informed children that the weblink would be shared with the consenting parent.

The *control group* received usual care. The intervention was offered to children in the control group after T2 measures were completed by the child and parent.

Data collection

Data collection occurred over three years (2015–2018). Participants in the intervention group completed T1 baseline and T2 post-intervention questionnaires. Parents were asked to complete a T3 intervention process survey at study end. Participants completed all measures electronically online through REDCap, a secure web-based application for building and managing online surveys and databases. The coordinator made reminder calls or sent reminder e-mails for surveys not completed within one week. This article presents T1 and T2 quantitative data related to child QOL. The median time between T1 and T2 was 69 days (range 7–176) for the children, and 68 days (range 7–176) for parents.

Measurement tools

Children and parents completed the 27-item *PedsQL 3.0 Cancer Module* to measure child QOL.¹⁹ We used the Child Self-Report versions for ages 5–7, 8–12, and 13–17. Parents completed the Parent Proxy-Report about their child. Children were asked to rate how much each item had been a problem over the past one month on a three-point (for children 7 years of age) or five-point (for children 8–17 years of age) Likert scale. The multidimensional scales include pain and hurt, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance, and communication. Scores for each dimension are reported. Higher scores indicate lower problems. Primary outcomes of interest included procedural and treatment anxiety, worry, cognitive problems, and communication dimensions of QOL. Previous studies show average internal consistencies for Cancer Module Scales of 0.72 for child report and 0.87 for parent report and are acceptable for group comparisons.

Analysis

Data analyses were conducted using IBM SPSS Statistics (Version 25). Frequency distributions were generated for summarizing the nominal and ordinal variables. Means and standard deviations were used to summarize the normally distributed continuous variable and medians and interquartile ranges were used to summarize the skewed distributions. Independent sample *t*-tests and chi-square tests of independence were conducted to compare the demographic and clinical characteristics of the participants in the two study data analysis groups. Comparisons between the study groups in the amount of change in each of the study outcome variables from baseline were conducted using generalized linear models that included the respective baseline score and time

between T1 and T2 as covariates. Cohen's *d* statistics quantifying the effect of the intervention on each of the outcome variables were generated by transforming the regression coefficient for the group effect after controlling for the baseline value. Interpretations of statistical significance maintained a maximum alpha of 0.05 ($p < 0.05$). Given that effect sizes were generated and presented, no adjustment for multiple outcomes to the critical alpha level was made.

Results

Participants

Of 2393 parents who self-identified as eligible after completing the electronic screening questions, 496 provided their basic demographic and personal information to be contacted (Fig. 2). We successfully contacted 273 parents. A total of 150 child-parent dyads (55%) enrolled and began participation in the study. Forty-two (28%) dyads dropped out before completing T2 (control=11 and experimental=31) and 10 (7%) completed T2 more than six months after enrollment (control=7 and experimental=3). One dyad was excluded due to eligibility concerns after finding an inconsistency on the parent-reported child date of birth. Therefore, 97 dyads were included in analysis. Summaries of the demographic and clinical characteristics of those completing the study and those excluded have been previously published.¹⁸ Region of the country and marital status were the only statistically significant differences observed ($p < 0.05$).

Other than home region, no statistically significant difference in demographic or clinical characteristics was ob-

served between the intervention and control groups of participants who completed the study (Tables 1 and 2). The sample of 97 ill children averaged 10 years of age. The majority were female ($n=57$, 59%) and white ($n=81$, 85%). Most had experienced a cancer relapse ($n=69$, 71%). Few had been notified that their cancer was terminal ($n=7$, 7%), a do-not-resuscitate order in place ($n=2$, 2%), received a hospice referral ($n=2$, 2%), or received a palliative care consult ($n=7$, 7%). The majority of caregivers were biological parents ($n=91$, 94%), female ($n=88$, 93%), Caucasian ($n=88$, 93%), living in the Midwest ($n=53$, 56%), married ($n=49$, 52%), and college educated ($n=64$, 67%), and had an annual family income of $> \$25,000$ ($n=53$, 56%) (Tables 1 and 2).

Descriptive summaries of the PedsQL scores at baseline (T1) and changes in those scores between T1 and end of study (T2) are shown in Table 3. Although not statistically significant, legacy-making demonstrated small effects for improvements in child procedural anxiety and child perceived physical appearance (Cohen's *d* 0.35–0.28, respectively) compared to the wait-list control group (Table 3).

Discussion

This study is the first of its kind to examine QOL effects of a web-based legacy intervention for children with advanced cancer. Previous studies have utilized face-to-face legacy intervention delivery mechanisms and shown promise to improve outcomes for patients with life-threatening conditions and their families.¹⁰ Our results are the first to support the feasibility of a RCT web-based legacy intervention.

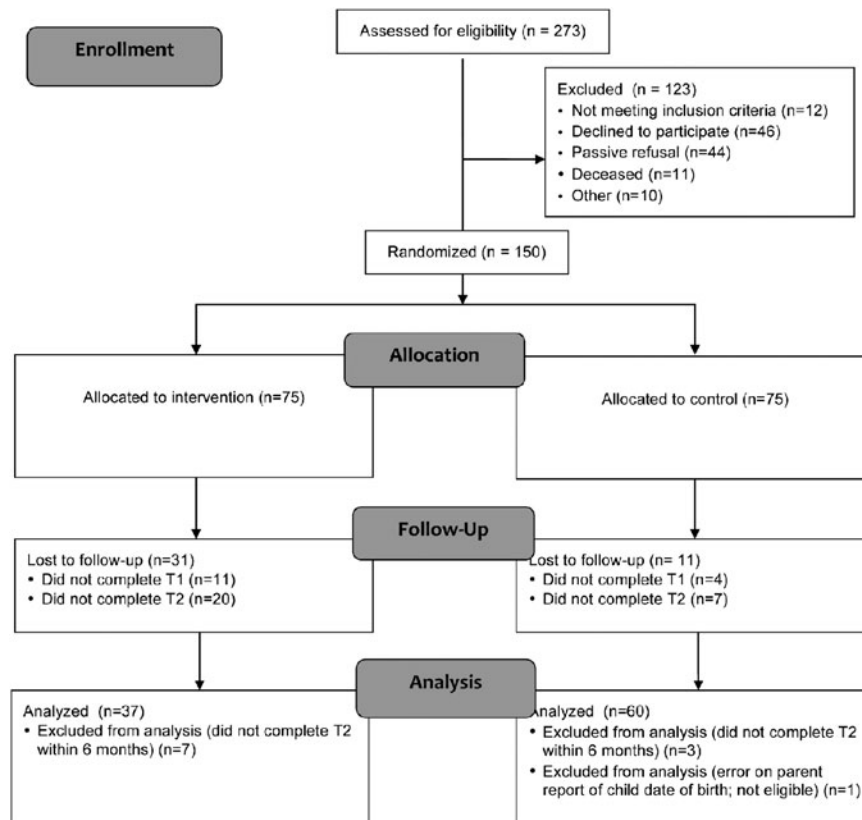


FIG. 2. CONSORT flow diagram.

TABLE 1. DEMOGRAPHIC CHARACTERISTICS BY STUDY GROUP (N=97)

<i>Characteristic</i>	<i>Overall (N=97)</i> <i>Mean (SD)</i> <i>N=94</i>	<i>Control (n=60)</i> <i>Mean (SD)</i> <i>n=57</i>	<i>Experimental (n=37)</i> <i>Mean (SD)</i>	<i>p</i>
<i>Child with Cancer</i>				
Age (years)	10.4 (3.0)	10.6 (3.0)	10.1 (3.0)	0.435
Gender				
Male	40 (41.2)	24 (40.0)	16 (43.2)	0.753
Female	57 (58.8)	36 (60.0)	21 (56.8)	
Race	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
White	81 (85.3)	51 (86.4)	30 (83.3)	0.059
Black or African American	4 (4.2)	4 (6.8)	0 (0.0)	
Asian	2 (2.1)	2 (3.4)	0 (0.0)	
American Indian or Alaska Native	2 (2.1)	1 (1.7)	1 (2.8)	
Other	6 (6.3)	1 (1.7)	5 (13.9)	
Ethnicity	<i>N=96</i>	<i>N=59</i>		
Hispanic or Latino	11 (11.5)	6 (10.2)	5 (13.5)	0.617
Not Hispanic or Latino	85 (88.5)	53 (89.8)	32 (86.5)	
Primary language				
English	95 (97.9)	59 (98.3)	36 (97.3)	0.727
Spanish	2 (2.1)	1 (1.7)	1 (2.7)	
Caregiver				
Relationship to child				
Biological parent	91 (93.8)	55 (91.7)	36 (97.3)	0.659
Adoptive parent	4 (4.1)	3 (5.0)	1 (2.7)	
Foster parent	1 (1.0)	1 (1.7)	0 (0.0)	
Grandparent	1 (1.0)	1 (1.7)	0 (0.0)	
Gender	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
Male	7 (7.4)	2 (3.4)	5 (13.9)	0.057
Female	88 (92.6)	57 (96.6)	31 (86.1)	
Race	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
White	88 (92.6)	54 (91.5)	34 (94.4)	0.238
Black or African American	4 (4.2)	4 (6.8)	0 (0.0)	
American Indian or Alaska Native	1 (1.1)	0 (0.0)	1 (2.8)	
Other	2 (2.1)	1 (1.7)	1 (2.8)	
Ethnicity	<i>N=90</i>	<i>N=55</i>	<i>N=35</i>	
Hispanic or Latino	5 (5.6)	3 (5.5)	2 (5.7)	0.958
Not Hispanic or Latino	85 (94.4)	52 (94.3)	33 (94.3)	
Home region	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
Northeast	12 (12.6)	10 (16.9)	2 (5.6)	0.019
Southeast	18 (18.9)	16 (27.1) ^a	2 (5.6) ^b	
Middle West	53 (55.8)	26 (44.1) ^a	27 (75.0) ^b	
Southwest	10 (10.5)	6 (10.2)	4 (11.1)	
West	2 (2.1)	1 (1.7)	1 (2.8)	
Primary language	<i>N=94</i>	<i>N=58</i>	<i>N=36</i>	
English	91 (96.8)	55 (94.8)	36 (100.0)	0.165
Spanish	3 (3.2)	3 (5.2)	0 (0.0)	
Highest grade completed	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
Grade school (K-8)	1 (1.1)	1 (1.7)	0 (0.0)	0.587
High school (9-12)	27 (28.8)	15 (25.4)	12 (33.3)	
GED	3 (3.2)	1 (1.7)	2 (5.6)	
College (undergraduate)	52 (54.7)	35 (59.3)	17 (47.2)	
Graduate school	12 (12.6)	7 (11.9)	5 (13.9)	
Current marital status	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
Never married	24 (25.3)	12 (20.3)	12 (33.3)	0.201
Married	49 (51.6)	32 (54.2)	17 (47.2)	
Divorced	11 (11.6)	7 (11.9)	4 (11.1)	
Separated	1 (1.1)	0 (0.0)	1 (2.8)	
Widowed	6 (6.3)	6 (10.2)	0 (0.0)	
Other	4 (4.2)	2 (3.4)	2 (5.6)	

(continued)

TABLE 1. (CONTINUED)

<i>Characteristic</i> <i>Child with Cancer</i>	<i>Overall (N=97)</i> <i>Mean (SD)</i> <i>N=94</i>	<i>Control (n=60)</i> <i>Mean (SD)</i> <i>n=57</i>	<i>Experimental (n=37)</i> <i>Mean (SD)</i>	<i>p</i>
Current annual family income	<i>N=95</i>	<i>N=59</i>	<i>N=36</i>	
Under \$25,000 per year	42 (44.2)	25 (42.4)	17 (47.2)	0.319
\$25,001–\$50,000 per year	20 (21.1)	11 (18.6)	9 (25.0)	
\$50,001–\$75,000 per year	11 (11.6)	7 (11.9)	4 (11.1)	
\$75,001–\$100,000 per year	11 (11.6)	10 (16.9)	1 (2.8)	
\$100,001 or more per year	11 (11.6)	6 (10.2)	5 (13.9)	

Superscripts (a,b) indicate specific regions accounting for the statistically significant difference, Bonferroni corrected, $p < 0.05$.

Enrollment was successfully achieved, and random assignment to the intervention or usual care group occurred as intended. Important discoveries resulted, including uneven attrition with the greater attrition in the intervention arm, meriting a strong focus on this in the future using qualitative

and quantitative methods with child and parent assessments. The study did not find convincing evidence supporting the hypothesis that legacy-making improved child dimensions of QOL across time. Overall, this is a null study that warrants discussion on possible reasons for limited findings.

TABLE 2. CLINICAL CHARACTERISTICS BY STUDY GROUP (N=97)

<i>Characteristic</i>	<i>Overall (N=97)</i> <i>N (%)</i>	<i>Control (N=60)</i> <i>N (%)</i>	<i>Experimental (N=37)</i> <i>N (%)</i>	<i>p</i>
Cancer relapse or recurrence				
No	28 (28.9)	16 (26.7)	12 (32.4)	0.543
Yes	69 (71.1)	44 (73.3)	25 (67.6)	
Secondary cancer				
No	95 (97.9)	58 (96.7)	37 (100.0)	0.262
Yes	2 (2.1)	2 (3.3)	0 (0.0)	
Surgery to remove tumor				
No	56 (57.7)	36 (60.0)	20 (54.1)	0.565
Yes	41 (42.3)	24 (40.0)	17 (45.9)	
Chemotherapy				
No	5 (5.2)	2 (3.3)	3 (8.1)	0.302
Yes	92 (94.8)	58 (96.7)	34 (91.9)	
Radiation				
No	28 (28.9)	18 (30.0)	10 (27.0)	0.754
Yes	69 (71.1)	42 (70.0)	27 (73.0)	
Bone marrow transplant	<i>N=96</i>		<i>N=36</i>	
No	83 (86.5)	50 (83.3)	33 (91.7)	0.248
Yes	13 (13.5)	10 (16.7)	3 (8.3)	
Phase I study				
No	63 (64.9)	37 (61.7)	26 (70.3)	0.395
Yes	14 (14.4)	8 (13.3)	6 (16.2)	
Unsure	20 (20.6)	15 (25.0)	5 (13.5)	
Notified cancer is terminal				
No	90 (92.8)	55 (91.7)	35 (94.6)	0.588
Yes	7 (7.2)	5 (8.3)	2 (5.4)	
DNR order in place				
No	95 (97.9)	60 (100.0)	35 (94.6)	0.069
Yes	2 (2.1)	0 (0.0)	2 (5.4)	
Hospice				
No	94 (96.9)	58 (96.7)	36 (97.3)	0.692
Yes	2 (2.1)	1 (1.7)	1 (2.7)	
Unknown	1 (1.0)	1 (1.7)	0 (0.0)	
Palliative care				
No	85 (87.6)	51 (85.0)	34 (91.9)	0.195
Yes	7 (7.2)	4 (6.7)	3 (8.1)	
Unknown	5 (5.2)	5 (8.3)	0 (0.0)	

DNR, do-not-resuscitate order.

TABLE 3. PEDSQL SUMMARIES

<i>Measure</i>	<i>Baseline Median [IQR]</i>	<i>Change Median [IQR]</i>	<i>p</i>	<i>Effect size^a</i>
PedsQL (child reports)				
Pain			0.411	-0.18
Control (<i>n</i> =56)	37.5 [15, 72]	0.0 [-13, 10]		
Intervention (<i>n</i> =32)	25.0 [0, 50]	0.0 [-13, 0]		
Nausea			0.298	-0.22
Control (<i>n</i> =57)	40.0 [20, 70]	0.0 [-18, 10]		
Intervention (<i>n</i> =33)	30.0 [0, 70]	0.0 [-15, 0]		
Procedural anxiety			0.176	0.28
Control (<i>n</i> =57)	33.3 [4, 67]	0.0 [-13, 9]		
Intervention (<i>n</i> =33)	25.0 [0, 55]	0.0 [0, 25]		
Treatment anxiety			0.958	0.01
Control (<i>n</i> =57)	50.0 [25, 80]	0.0 [-9, 9]		
Intervention (<i>n</i> =33)	25.0 [0, 75]	0.0 [0, 9]		
Worry			0.987	0.01
Control (<i>n</i> =57)	33.3 [12, 67]	10.0 [9, 11]		
Intervention (<i>n</i> =33)	25.0 [0, 59]	10.0 [9, 11]		
Cognition			0.384	-0.18
Control (<i>n</i> =57)	30.0 [22, 55]	3.0 [2, 4]		
Intervention (<i>n</i> =33)	30.0 [0, 70]	3.0 [2, 3]		
Perceived physical appearance			0.116	0.35
Control (<i>n</i> =57)	50.0 [25, 80]	1.0 [0, 2]		
Intervention (<i>n</i> =33)	25.0 [0, 75]	1.0 [1, 2]		
Communication			0.821	0.05
Control (<i>n</i> =57)	41.7 [25, 71]	0.0 [-17, 17]		
Intervention (<i>n</i> =33)	50.0 [0, 75]	0.0 [0, 9]		
PedsQL (parent proxy reports)				
Pain			0.274	-0.23
Control (<i>n</i> =58)	37.5 [9, 63]	0.0 [-13, 13]		
Intervention (<i>n</i> =34)	25.0 [0, 54]	0.0 [13, 13]		
Nausea			0.055	-0.42
Control (<i>n</i> =58)	42.5 [12, 62]	0.0 [-5, 15]		
Intervention (<i>n</i> =34)	25.0 [0, 59]	0.0 [-25, 10]		
Procedural anxiety			0.963	0.01
Control (<i>n</i> =58)	37.5 [0, 75]	0.0 [-11, 17]		
Intervention (<i>n</i> =33)	25.0 [0, 50]	0.0 [-9, 9]		
Treatment anxiety			0.209	-0.27
Control (<i>n</i> =58)	41.7 [22, 69]	0.0 [-9, 17]		
Intervention (<i>n</i> =34)	25.0 [0, 80]	0.0 [-25, 11]		
Worry			0.733	-0.07
Control (<i>n</i> =58)	33.3 [16, 61]	0.0 [-11, 11]		
Intervention (<i>n</i> =33)	25.0 [0, 63]	0.0 [-9, 9]		
Cognition			0.428	0.16
Control (<i>n</i> =58)	35.0 [17, 64]	0.0 [-6, 7]		
Intervention (<i>n</i> =34)	25.0 [0, 65]	0.0 [-20, 17]		
Perceived physical appearance			0.607	0.11
Control (<i>n</i> =58)	50.0 [25, 75]	0.0 [-17, 9]		
Intervention (<i>n</i> =33)	25.0 [0, 80]	0.0 [-25, 25]		
Communication			0.949	0.01
Control (<i>n</i> =58)	45.8 [14, 84]	0.0 [-13, 9]		
Intervention (<i>n</i> =34)	37.5 [0, 84]	0.0 [-17, 17]		

^aCohen's *d* estimate transformed from regression coefficient, after controlling for baseline values and the length of time between T1 and T2.

IQR, interquartile ranges.

We found no statistically significant effect of the intervention for any of the primary QOL outcomes (procedural and treatment anxiety, worry, cognitive problems, and communication dimensions). This contrasted parent reports on the T3 satisfaction survey that the intervention helped children express their feelings (86.4%) and feel better emotionally (59.3%).¹⁸ This discrepancy between quantitative and qualitative results has been seen in our previous work as well as on other main outcomes of this study reported elsewhere.^{15,20} Previous studies have suggested that legacy interventions may improve emotional dimensions of QOL and communication.^{12,15} The PedsQL may not be the best measure to capture effects of our intervention, and QOL as operationalized by the PedsQL may not be the exact outcome influenced by our intervention. In addition, intervention content or delivery format could have eroded effectiveness. Future work is needed, using both qualitative and quantitative methods, to determine if and how the intervention could improve components of child QOL.

Raw change scores suggest the intervention had little effect on child dimensions of QOL, with the notable exception of child worry, which increased for both the intervention and control groups based on child self-reports, but not parent proxy reports. Parent-child communication is not only a key area for future research but also a key area of refinement needed for our intervention based on parent feedback that the intervention served as a communication tool and positive intervention effects on child-father communication.²⁰ The discrepancy between child and parent proxy reports regarding worry highlights the need for pediatric research to involve multiple informants, including children, mothers, and fathers. While it is not uncommon for pediatric palliative care studies to sample child-parent dyads,^{2,21} or mothers and fathers of seriously ill children,²² studies rarely have included reports directly from all three: ill children, mothers, and fathers, something critical to implementing effective family-centered palliative care interventions.

Limitations

A major study limitation is the difference in participant numbers between intervention and control groups. Attrition in the intervention group could have been due, in part, to issues with user friendliness of the intervention web-program (e.g., difficulties logging into website). Generalizability of our results is limited to children 7 to 17 years of age with relapsed/refractory cancer recruited through Facebook. Outcomes could have been affected by varied cancer diagnoses. We acknowledge that the control group received “usual care,” yet usual care may have varied across participants recruited from all over the United States, and no data were collected on palliative care interventions children may have been receiving. The large discrepancy of number of days between T1 and T2 assessment points could have affected study outcomes. Despite these limitations, this study contributed new knowledge to the science in pediatric palliative care and demonstrated promising areas for future legacy-focused research studies.

Implications for research and practice

Future research should examine potential moderators (e.g., age and gender) of legacy effects and which populations are

most likely to benefit. It is possible that the intervention was effective for only certain subgroups of our sample (e.g., teenagers and females). We are currently revising the intervention protocol and measurement tools based on study results, including parent feedback. Further work will be needed to determine feasibility and acceptability of the revised intervention, and then a subsequent pilot study to examine preliminary efficacy to determine if a larger RCT is warranted. The increased worry over time in both groups is noteworthy and has implications for future intervention studies, including the consideration of a certain extent of increase being factored into an analysis. Providers can help share with families that children with life-threatening conditions are willing and able to participate in legacy activities. Palliative care providers are in ideal roles to help facilitate conversations with families that children with advanced illnesses have already created legacies, but may benefit from documenting their legacies in some way.

Conclusion

Our study resulted in limited evidence to support the hypothesis that our web-based legacy intervention improved dimensions of QOL for children with cancer. Whether the intervention was simply lacking in effect (intervention), it was delivered in a manner that eroded effectiveness (design), quantitative measurement tools did not capture its effects (measurement), or participants did not report sensitively (reporters), cannot be determined. Qualitative parent feedback provides strong evidence that the intervention has promise to improve outcomes for children with advanced cancer; thus, further revision of the intervention and measurement protocol is warranted. More work is needed to determine how legacy interventions can be used to improve QOL in pediatric palliative care.

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Address correspondence to:
 Terrah Foster Akard, PhD, RN, CPNP, FAAN
 Vanderbilt University School of Nursing
 and School of Medicine
 461 21st Avenue South
 418 Godchaux Hall
 Nashville, TN 37240
 USA

E-mail: terrah.akard@vanderbilt.edu