Childhood Cancer Survivors' Knowledge About Their Past Diagnosis and Treatment

Childhood Cancer Survivor Study

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Context Adult survivors of childhood cancer are at risk for adverse effects later in life but may have limited access to information about their diagnosis and treatment. This knowledge is necessary to motivate them to seek medical follow-up and to report essential history to health care professionals.

Objective To assess knowledge of adult survivors of childhood cancer about their primary cancer diagnosis and associated therapies.

Design, Setting, and Participants Cross-sectional survey of 635 consecutive survivors (approximately 5%) drawn from 12 156 participants 18 years or older participating in the Childhood Cancer Survivor Study (a multiinstitutional cohort of individuals diagnosed between January 1, 1970, and December 31,1986, at an age <21 years, who had survived 5 years from diagnosis).The survey assessed knowledge of their cancer diagnosis and associated therapies in a 3- to 5-minute telephone questionnaire.

Main Outcome Measures Responses were compared with medical record data for accuracy, sensitivity, specificity, and positive and negative predictive value.

Results Overall, 72% accurately reported their diagnosis with precision and 19% were accurate but not precise. Individuals with central nervous system (CNS) cancer (odds ratio , 5.1; 95% confidence interval , 2.6-9.9) and neuroblastoma (OR, 4.2; 95% CI, 1.8-9.6) were more likely not to know their cancer diagnosis. Participants' accuracy rates for reporting their treatment history was 94% for chemotherapy, 89% for radiation, and 93% for splenectomy. Among those who received anthracyclines, only 30% recalled receiving daunorubicin therapy and 52% recalled receiving doxorubicin therapy, even after prompting with the drugs' names. Among those who received radiotherapy, 70% recalled the site of radiotherapy. History of receiving a written medical summary, attending a long-term follow-up clinic, and anxiety about late effects were not associated with greater knowledge.

Conclusions Important knowledge deficits exist among adult survivors of childhood cancer regarding basic aspects of their diagnosis and treatment. Such deficits could impair survivors' ability to seek and receive appropriate long-term follow-up care.

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CHILDHOOD CANCER CURE RATES have increased dramatically over the past few decades, with overall 5-year survival rates now exceeding 70%.[1] This growing population of survivors is at risk for adverse effects related to their malignancy and subsequent therapy. These risks include second neoplasms, organ dysfunction, early death, endocrine abnormalities, and neuropsychological dysfunction.[2-7] Survivors may benefit from anticipatory guidance and ongoing surveillance to minimize morbidity and mortality.[8] The survivor must have adequate knowledge of his/her cancer diagnosis and treatment to be motivated to pursue necessary medical follow-up and to relate accurately and completely his/her medical history to health care professionals.

Unlike their adult counterparts, childhood cancer survivors probably had limited access to information pertaining to their malignancy at the time of their diagnosis and treatment. They may have been too young to understand explanations regarding the disease and treatments. Their parents may have decided to shield them from details of their disease, including terms such as "cancer" and "chemotherapy." Also, parents usually take

See also Patient Page.

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responsibility for making decisions affecting their children, such as consenting to procedures and therapy. Thus, survivors of childhood cancer may be unable to recall relevant information about their cancer medical history.

Nested within an ongoing cohort study, we conducted a cross-sectional study of 635 childhood cancer survivors who were diagnosed from 1970 through 1986 to determine the accuracy, sensitivity, specificity, and predictive value of self-reported information about their primary cancer diagnosis and treatments compared with medical records. We hypothesized that a lower level of knowledge would be associated with the following: younger age at cancer diagnosis, diagnosis during an earlier treatment era, history of head or neck radiation, limited clinical follow-up, fewer years of formal education, younger current age, no history of a subsequent malignancy, and less concern about potential late effects.

METHODS

Subjects

The Childhood Cancer Survivor Study (CCSS) is a multi-institutional study of individuals at 25 clinical centers who have survived at least 5 years after diagnosis of "a cancer, leukemia or similar illness during childhood." The study design and cohort characteristics have been described previously.[2,4,5,9] Inclusion criteria for the CCSS cohort are (1) diagnosis of leukemia, central nervous system (CNS) tumor, Hodgkin disease, non-Hodgkin lymphoma, malignant kidney tumor, neuroblastoma, soft tissue sarcoma, or bone tumor (list of all eligible *International Classification of Diseases* (*ICD*) O codes within diagnosis categories can be found at www.cancer.umn.edu/ccss); (2) diagnosis and initial treatment at one of the collaborating centers between January 1, 1970, and December 31, 1986; (3) age younger than 21 years at diagnosis; and (4) survival of more than 5 years after diagnosis. Of the 20276 eligible patients, 14054 (69.0%) participated; 2996 (14.8%) were lost to follow-up; 3132 (15.4%) refused participation; and 94 (<1%) are pending return of a baseline questionnaire. Of the 14054 participants, 12156 were both alive and at least 18 years old on April 1, 2001.

Beginning August 1, 1994, participants completed an extensive baseline questionnaire about demographic characteristics, socioeconomic status, health status, health behaviors, and family history. Trained data abstractors at the treating institutions reviewed participants' medical records to ascertain cancer diagnosis and treatment information. Recorded data included qualitative and quantitative chemotherapy information, fields of radiation therapy, and types of surgery performed. All radiation records were reviewed separately by a CCSS radiation oncologist for accuracy and validity. Details of the baseline survey and medical record abstraction form used in data collection are available at www.cancer.umn.edu/ccss. Details regarding the coding of medical records and interevaluator reliability have been described.[9]

After a median of 5.2 years since the baseline survey, a follow-up questionnaire (starting May 1, 2000) was being administered to all the members of the cohort to update data related to their survivor experience. In our study, an unselected consecutive series of 643 living CCSS participants, also at least 18 years old, who were successfully contacted for the follow-up survey by 5 selected telephone interviewers underwent an additional 3- to 5-minute telephone questionnaire prior to the interview. The goal was to sample 5%. A priori, the maximum tolerable widths of 95% confidence intervals (CIs) for the respondents' knowing their diagnosis were calculated to be about plus or minus 3.5% when the true fraction of correct answers was 75% (the mid point between a random guess, 50%, and a perfect guess, 100%). This required a minimum sample size of 588, which was rounded-up to 5% of the cohort (n=625).

Of the 643 who were contacted, 6 respondents were excluded because of input by another individual during the telephone call, and 2 of the respondents contacted for the follow-up survey refused this ancillary study. The remaining 635 participants were representative of the entire CCSS cohort. (TABLE 1). Nine individuals had not returned a signed medical release allowing access to their medical record data, thereby precluding analysis of their therapy data.

All CCSS protocol and contact documents were reviewed and approved by the human subjects committee at the University of Minnesota and at each participating institution if required.

Case Knowledge Ascertainment

In the supplemental telephone questionnaire (BOX), respondents were asked to recall the name of their "cancer, leukemia, tumor, or similar illness of childhood" and whether their treatment included chemotherapy, radiation therapy, or surgery. The participant was asked probing questions to provide the most detailed response known (eg, the subtype of leukemia or lymphoma or the histology of the brain tumor). If any of the therapy questions was answered affirmatively, participants were asked to spontaneously recall the names of any chemotherapeutic agents, sites of radiation therapy, and/or types of surgical procedures. Those who reported having received chemotherapy were asked directly if they remembered receiving daunorubicin (or daunomycin) or doxorubicin (or adriamycin). Additionally, participants were asked if they believed that cancer treatment could cause serious future health problems. After conducting a pilot of 110 individuals, 2 additional questions regarding clinic attendance and possession of a treatment summary were added to the end of the questionnaire. Individuals in the pilot had a slightly greater proportion of patients with leukemia (46% vs 35%) but were similar to the other participants in terms of age at diagnosis, era of diagnosis, and therapy received.

Responses from the previously completed baseline questionnaire were assessed to determine the participants'

Table 1. Comparison of Study Sample to Nonparticipating Members of the Childhood Cancer Survivor Study (CCSS) Cohort*

Characteristics	Participating Sample	Nonparticipating Cohort*	*P* Value†
No. of cases	635	11 521	
Women, No. (%)	295 (46)	5402 (47)	.83
Diagnosis, No. (%)			
Leukemia	221 (35)	3895 (34)	.07
Central nervous system tumors	82 (13)	1427 (12)	
Hodgkin disease	86 (14)	1582 (14)	
Non-Hodgkin lymphoma	53 (8)	931 (8)	
Wilms tumor	65 (10)	988 (9)	
Neuroblastoma	47 (7)	682 (6)	
Soft tissue sarcoma	35 (6)	1031 (9)	
Bone tumors	46 (7)	985 (8)	
Age at diagnosis, y			
Mean (SD)	7.9 (5.5)	8.0 (5.8)	.67
Median (range)	7 (0-20)	7 (0-20)	
Current age, y			
Mean (SD)	29.3 (6.8)	29.3 (7.2)	.78
Median (range)	29 (18-49)	29 (18-51)	
Elapsed time since diagnosis, y			
Mean (SD)	20.9 (4.4)	20.9 (4.5)	.79
Median (range)	21 (14-31)	20 (14-31)	

*Nonparticipating cohort members who were alive and at least 18 years as of April 1, 2001.
†Nonparticipating but eligible CCSS cohort compared with participating study sample by χ^2 test for categorical variables and *t* test for continuous variables.

Box. Patient Knowledge Telephone Survey

1. Name of primary cancer, leukemia, or similar illness of childhood
   Is there a more specific name?
   If leukemia: acute lymphoblastic leukemia, acute myelogenous leukemia, or chronic myelogenous leukemia?
   If lymphoma: Hodgkin or non-Hodgkin disease?
2. Were chemotherapy, radiation therapy, and surgery administered? (yes/no/don't know)
3. Names of chemotherapeutic agents if "yes" to chemotherapy.
4. Sites of radiation therapy if "yes" to radiotherapy.
5. Types of surgical procedures if "yes" to surgery.
6. If "yes" or "don't know" to chemotherapy, was anthracycline therapy administered?
   Daunorubicin or daunomycin (yes/no/don't know)
   Doxorubicin or adriamycin (yes/no/don't know)
7. Do you feel that previous treatment could cause serious future health problems? (yes/no/don't know)

Questions Not Asked of Pilot Study Participants

8. Have you ever attended a clinic for the purpose of late effects follow-up? (yes/no/don't know)
9. Have you ever received a written summary of disease and treatment? (yes/no/don't know)

self-reported demographic information (age, sex, education, income) and history of cardiac complications (cardiac symptoms, consultation of a cardiologist, etc).

Data Analysis

Each participant's responses to the cancer and treatment history supplemental questions were compared with his or her medical record. Accuracy of diagnosis report was assigned to 1 of 5 categories based on the detail of the subject's best response: (1) accurate with detail, (2) accurate without detail, (3) "cancer" or "tumor" with no further knowledge, (4) incorrect, and (5) unknown. Examples of reports that were considered accurate without detail included recalling a diagnosis of "lymphoma," but not distinguishing correctly between Hodgkin and non-Hodgkin lymphoma. All reports of "brain tumor" were classified as accurate without detail, as were 7 cases of astrocytoma and juvenile pilocytic astrocytoma reported as a "benign brain tumor." A self-report of "lymphoblastic leukemia" for "lymphoblastic lymphoma" was considered accurate with detail because of changing diagnostic criteria with respect to bone marrow involvement.

Sensitivity and specificity of self-reported cancer diagnosis, history of chemotherapy, anthracycline therapy, radiation therapy, and splenectomy were calculated. We focused on splenectomy because it is a surgical procedure with important considerations for long-term follow-up. Participant accuracy, sensitivity, specificity, positive predictive value, and negative predictive value were also stratified by host characteristics of interest, which included the following: type of cancer; age at diagnosis (<5 years or ≥5 years); era of cancer diagnosis (1970-1977 or 1978-1986); current age (18-24 years, 25-34 years, or ≥35 years); educational level (less than high school diploma, high school diploma but not college degree, or college degree); history of relapse, second primary malignancy, or radiation to the head and neck region; worry about future health problems; history of ever seeking long-term follow-up care; and history of receiving written summary of treatment. The statistics, sensitivity and specificity, in this analysis were used as performance summaries for a binary estimate (ie, guess, prediction, or decision) in relation to the true binary status.

Likelihood-based analyses of odds ratios (ORs), with exact inference methods when necessary, were used to examine the associations between in-

correctly reporting aspects of the cancer medical history and the various survivor attributes of interest. We also tested whether the association between diagnosis before the age of 5 years and knowledge varied with the era of cancer diagnosis. Multiple logistic regression models were constructed to include all of the covariates that were significantly associated with inaccurate reporting in univariate analysis. Colinearity was assessed by cross-tabulations and plots of the analyzed covariates before proceeding to multiple logistic regression. No appreciable colinearity was observed. Data were analyzed with the SAS PC software package version 6.12 (SAS institute, Cary, NC) with 2-tailed statistical tests.

RESULTS

Report of Diagnosis

When prompted with choices of names of different diagnoses, 72% of the participants accurately reported their diagnosis with detail and 19% were accurate without detail (TABLE 2). These percentages varied by cancer category. Only 75% of individuals who had CNS cancers gave an accurate response. Ninety-eight percent of those with Hodgkin disease, Wilms tumor, and bone cancers were able to name their cancer diagnosis with detail.

We found no consistent reporting pattern among those who reported an incorrect diagnosis. The 7 incorrect responses by those who survived CNS cancer were "adenoma," "something with an A," "tumor sclerosis," "malignoma" "natural blastoma," "tuberous sclerosis," and "neurofibromatosis." Two of the 6 survivors of neuroblastoma who were incorrect said they had a history of leukemia. One Hodgkin disease survivor said he had non-Hodgkin lymphoma. One survivor of acute myelogenous leukemia reported a history of acute lymphoblastic leukemia. None of these incorrect reports occurred in those with a history of a second malignancy (n=14).

Without additional prompting, the proportion of responses that were accurate with detail would have been lower among those with leukemia (53% vs 61% with prompting), Hodgkin disease (67% vs 97%), and CNS cancer (24% vs 34%). Prompting resulted in minimal change in the proportion of those who gave inaccurate responses.

Diagnosis during an earlier treatment era, history of a CNS cancer, history of a neuroblastoma, and male sex were significantly associated with not knowing one's cancer diagnosis in unadjusted analysis (TABLE 3). Relative to all other cancers combined, a history of Hodgkin disease was associated with an increased likelihood of correctly recalling diagnosis. In multiple regression analysis, a history of CNS cancer was the strongest predictor of not knowing the name of one's cancer (OR, 5.1; 95% CI, 2.6-9.9).

Chemotherapy History

Among all participants, 94% accurately stated whether they had chemotherapy, 3% gave a wrong response, and 3% responded that they did not know (TABLE 4). Of those who provided a response, 10% falsely reported receiving chemotherapy when they had not. One percent did not report their chemotherapy when they had received it. Individuals with CNS cancer had the lowest likelihood (84%) of knowing their chemotherapy history.

In unadjusted analysis, diagnosis before the age of 5 years, diagnosis during an earlier treatment era, and a history of CNS malignancy were associated with not knowing one's chemotherapy history (Table 3). All those with non-Hodgkin lymphoma and soft tissue sarcoma were aware that they had undergone chemotherapy. After adjusting simultaneously for all the variables significant in univariate analysis, diagnosis at younger age at (OR, 2.8; 95% CI, 1.3-5.8), diagnosis during an earlier treatment era (OR, 2.3; 95% CI, 1.1-4.6), and history of a CNS cancer (OR, 3.6; 95% CI, 1.5-8.0) remained associated with not knowing one's chemotherapy history.

When asked to list their chemotherapy drug treatment, doxorubicin, 61 (33%) of 185 respondents recalled receiving doxorubicin vs 6 (8%) of 81 participants who received daunorubicin. When prompted with the drugs' names, 15% of 188 who had received doxorubicin and 18% of 81 who had received daunorubicin said that they did not know whether they had received these drugs. Among those who responded affirma-

Table 2. Patient Report of Previous Cancer Diagnosis by Category

Variables	No. of Patients	No. (%)						
		Accurate Responses			Inaccurate Responses			
		With Detail	Without Detail	Total	Cancer	Incorrect	Did Not Know	Total
Leukemia	221	134 (61)	74 (33)	208 (94)	0 (0)	2 (1)	11 (5)	13 (6)
Central nervous system cancer	82	28 (34)	34 (41)	62 (75)	7 (9)	7 (9)	6 (7)	20 (25)
Hodgkin disease	86	83 (97)	1 (1)	84 (98)	0 (0)	1 (1)	1 (1)	2 (2)
Non-Hodgkin lymphoma	53	38 (72)	9 (17)	47 (89)	1 (2)	5 (9)	0 (0)	6 (11)
Wilms tumor	65	63 (97)	1 (1)	64 (98)	0 (0)	0 (0)	1 (2)	1 (2)
Neuroblastoma	47	36 (77)	1 (2)	37 (79)	3 (6)	6 (13)	1 (2)	10 (21)
Soft tissue sarcoma	35	27 (78)	4 (11)	31 (89)	1 (3)	2 (5)	1 (3)	4 (11)
Bone	46	45 (98)	0 (0)	45 (98)	0 (0)	0 (0)	1 (2)	1 (2)
All diagnoses	635	454 (72)	124 (19)	578 (91)	12 (2)	23 (4)	22 (3)	57 (9)

tively or negatively, 52% of 163 who received doxorubicin and 30% of 66 who received daunorubicin could recall these particular therapies. Asking about the drugs by name reduced the positive predictive value of affirmative responses compared with those obtained by spontaneous recall from 94% to 75% for doxorubicin and 58% to 46% for daunorubicin. Individuals who had a positive self-reported family cardiac disease history had slightly more accurate reporting of anthracycline history than those with a negative family history (54% vs 45% for doxorubicin; 32% vs 25% for daunorubicin, respectively), when asked about these drugs by name.

Radiation History

Eighty-nine percent of the respondents accurately recalled whether they had received radiation therapy, 3% gave a wrong response, and 8% responded that they did not know. Ten percent of those who had not received radiation therapy thought they had; 1% of individuals who had radiation therapy were not aware of their history. Accuracy rates of survivors of neuroblastoma was 77%; Wilms tumor, 83%; and leukemia, 85% vs an accuracy rate of 99% among those with Hodgkin disease. Fifteen percent of those with neuroblastoma, 11% with Wilms tumor, and 11% with leukemia reported that they did not know whether they had radiation therapy.

Table 3. Multiple Logistic Regression Analysis of Inaccurate Reporting of Diagnosis, Chemotherapy, and Radiation History*

	Odds Ratio (95% Confidence Interval)					
	Diagnostic Report		Chemotherapy History		Radiation History	
Variables	Unadjusted	Adjusted†	Unadjusted	Adjusted†	Unadjusted	Adjusted†
Age, y						
At diagnosis						
<5	1.2 (0.7-2.1)	. . .	2.6 (1.3-5.3)‡	2.8 (1.3-5.8)‡	4.8 (2.7-8.3)§	2.0 (1.0-3.8)‖
≥5, referent	1.0	1.0	1.0	1.0	1.0	1.0
At interview						
18-24	0.7 (0.3-1.5)	. . .	1.1 (0.5-2.7)	. . .	13 (3.8-42.0)§	3.5 (0.8-15.0)
25-34	0.9 (0.5-1.8)	. . .	0.7 (0.3-1.7)	. . .	4.7 (1.4-16.0)	2.7 (0.8-9.5)
≥35, referent	1.0	1.0	1.0	1.0	1.0	1.0
Year diagnosed						
1970-1977	2.4 (1.4-4.2)‡	2.7 (1.5-4.9)§	2.1 (1.1-4.2)‖	2.3 (1.1-4.6)‖	0.5 (0.3-1.1)	. . .
1978-1986, referent	1.0	1.0	1.0	1.0	1.0	1.0
Diagnosis¶						
Leukemia	0.5 (0.3-1.1)	. . .	0.5 (0.2-1.2)		1.6 (0.9-2.7)	. . .
Central nervous system cancer	4.5 (2.4-8.2)§	5.1 (2.6-9.9)§	3.2 (1.4-6.9)‡	3.6 (1.5-8.0)‡	0.5 (0.2-1.4)	. . .
Hodgkin disease	0.2 (0.1-0.9)‡	0.4 (0.1-1.6)	1.4 (0.5-3.4)	. . .	0.1 (0.0-0.6)‖	0.2 (0.0-1.8)
Non-Hodgkin lymphoma	1.3 (0.5-3.2)	. . .	0.0 (0.0-0.6)	0.0 (0.0-1.2)	0.9 (0.3-2.3)	. . .
Wilms tumor	0.1 (0.0-1.1)	. . .	1.2 (0.4-3.6)	. . .	1.5 (0.7-3.2)	. . .
Neuroblastoma	3.1 (1.5-6.6)‡	4.2 (1.8-9.6)§	2.2 (0.8-6.1)	. . .	2.9 (1.4-6.0)‡	1.6 (0.8-3.7)
Soft tissue sarcoma	1.3 (0.4-3.8)	. . .	0.0 (0.0-0.9)‖	0.0 (0.0-1.1)	0.2 (0.0-1.7)	. . .
Bone cancer	0.2 (0.0-1.6)	. . .	0.4 (0.1-2.9)	. . .	0.8 (0.29-2.4)	. . .
Educational level						
≤11th grade	1.8 (0.8-4.3)	. . .	2.4 (0.9-6.9)	. . .	16.0 (3.9-70.0)§	6.7 (1.4-33.0)‡
High school diploma, some college	2.2 (1.0-5.0)	. . .	1.6 (0.5-4.4)	. . .	8.8 (2.1-37.0)	6.4 (1.5-28.0)
≥College degree, referent	1.0	1.0	1.0	1.0	1.0	1.0
Men	2.0 (1.1-3.6)‖	2.1 (1.1-3.9)‖	1.4 (0.7-2.9)	. . .	1.5 (0.9-2.5)	. . .
History of radiation to head or neck	0.9 (0.6-1.7)	. . .	1.8 (0.8-3.6)	. . .	0.3 (0.2-0.5)	. . .
Not worried about health	1.4 (0.1-2.7)	. . .	1.0 (0.5-2.2)	. . .	2.0 (1.0-3.9)‖	1.4 (0.7-2.5)
Did not receive clinical summary	0.9 (0.4-2.0)	. . .	1.6 (0.4-5.3)	. . .	1.2 (0.5-2.8)	. . .
No history of attending long-term follow-up clinic	1.0 (0.8-1.3)	. . .	2.2 (0.9-5.0)	. . .	1.8 (0.9-3.3)	. . .
History of recurrence	0.5 (0.2-1.6)	. . .	1.3 (0.4-3.8)	. . .	0.6 (0.2-1.6)	. . .
History of second malignancy	0.0 (0.0-1.5)	. . .	3.0 (0.6-14.0)	. . .	0.0 (0.0-1.2)	. . .
Income <$20 000/y	1.4 (0.7-2.7)	. . .	1.3 (0.5-3.0)	. . .	1.4 (0.7-2.6)	. . .

*Ellipses indicate that variables were not statistically significant in the univariate analysis and were thus excluded from the multiple logistic regression analysis. Inaccurate refers to respondents with cancer who reported nothing more than that they had cancer, remembered incorrectly, or answered that they did not know whether they had cancer. Similarly, responses of "do not know" to chemotherapy and radiation history were analyzed as inaccurate.

†The Hosmer and Lemeshow goodness-of-fit test was performed to assess the multiple regression models. For the outcomes of diagnosis knowledge ($P = .57$), chemotherapy knowledge ($P = .24$), and radiation knowledge ($P = .88$), goodness of fit was supported.

‡$P<.01$.

§$P<.001$.

‖$P<.05$.

¶Each diagnostic category covariate uses as a referent the remaining 7 diagnostic categories.

Age younger than 5 years at diagnosis, younger age at interview, history of neuroblastoma, lower level of education, and less concern regarding future health risks were associated with less awareness of history of radiation therapy (Table 3). A history of Hodgkin disease was associated with correctly reporting radiation history. After adjusting for age at interview, diagnosis of Hodgkin disease, and diagnosis of neuroblastoma, individuals diagnosed at a younger age (OR, 2.0; 95% CI, 1.0-3.8) and with less than a high school diploma (OR, 6.7; 95% CI, 1.4-33.0) vs those with at least a college degree were less likely to know their radiation therapy history.

The ability to recall the general location of radiation therapy was 70% overall but varied by the treatment site. Accurate reports were received among 73% who received head or neck radiation, 62% who received spine radiation, 64% who received chest radiation, 67% who received abdomen or pelvis radiation, 81% who received limb radiation, and 75% who received total body radiation. Twenty-five individuals (8%) who had not received radiation to the head or neck erroneously stated that they had. Those who overreported radiation history to the head or neck had the following diagnoses: leukemia (n=6), CNS cancer (n=10), Hodgkin disease (n=4), non-Hodgkin lymphoma (n=5).

Splenectomy History

Sixty-seven percent of all respondents with splenectomy correctly responded when they were asked without prompting to list all their previous surgeries, similar to the rate (69%) among those who had Hodgkin disease. However, 14% of those who had Hodgkin disease incorrectly recalled they had had a splenectomy when they did not have one.

Analysis of potential factors associated with not knowing one's history of splenectomy was limited to individuals who had Hodgkin disease (58 of the 63 splenectomies were performed in patients with this diagnosis). No other risk factors were found to be significantly associated with splenectomy knowledge by multiple logistic regression.

Health Beliefs and Practices

When asked if past therapies could cause a serious health problem with the passage of time, 35% of participants responded affirmatively; 46% responded negatively; and 19% did not know. Forty-four percent of respondents stated that they had attended a clinic expressly for follow-up of their cancer. Only 15% responded that they ever received a written list of their disease diagnoses and treatment to keep as a reference in the future. Interestingly, 12% of cases did not know if they had received such a summary. Neither attendance at a long-term follow-up clinic nor receipt of a written clinical summary was associated with greater awareness of one's diagnosis and treatment (Table 3).

COMMENT

Given the remarkable improvement in childhood cancer survival rates, the key objective of this study was to identify and characterize an issue that would affect the health and quality of life of long-term survivors. Accurate information regarding an individual's cancer diagnosis and treatment is necessary to provide appropriate follow-up health care. Therefore, our findings need to be translated into interventions that will modify any deficiencies. This study provides evidence that knowledge deficits exist among adult survivors of childhood cancer about ba-

Table 4. Accuracy, Sensitivity, Specificity, and Predictive Value of Reported History of Chemotherapy, Radiation Therapy, and Splenectomy*

		No./Total (%) Who Responded				
		Chemotherapy History				
Variable	Definition	Overall	Daunorubicin‡	Doxorubicin‡	Radiation History	Splenectomy History
Accuracy	Correct	592/626 (94)	533/626 (85)	482/626 (77)	560/626 (89)	584/626 (93)
	Incorrect	18/626 (3)	77/626 (12)	128/626 (20)	18/626 (3)	25/626 (4)
	Do not know†	16/626 (3)	16/626 (3)	16/626 (3)	48/626 (8)	17/626 (3)
Sensitivity	Proportion of sample who responded affirmatively among those who received this therapy	479/484 (99)	6/81 (8)	61/185 (33)	400/401 (99)	42/63 (67)
Specificity	Proportion of sample who responded negatively among those who did not receive this therapy	113/126 (90)	527/529 (99)	421/425 (99)	160/177 (90)	542/546 (99)
Positive predictive value	Proportion of sample who received this therapy among those who responded affirmatively	479/492 (97)	6/8 (75)	61/65 (94)	400/417 (96)	42/46 (91)
Negative predictive value	Proportion of sample who did not receive this therapy among those who responded negatively	113/118 (96)	527/602 (88)	421/545 (77)	160/161 (99)	542/563 (96)

*Ascertained from unprompted report of all surgeries performed.
†Those who responded "do not know" were not included in calculations of sensitivity, specificity, and predictive value.
‡Ascertained from unprompted report of all chemotherapy received.

sic facts of their diagnosis and treatment. Many individuals who were interviewed were unable to report the necessary elements of an adequate medical history.

Depending on the situation, both overreporting and underreporting, can have deleterious implications. Only 74% could provide an accurate *general* summary of all of the elements of their cancer history. No one could provide an accurate *detailed* summary, ie, the detailed name of the cancer, whether doxorubicin or daunorubicin was administered, and the site of any radiation therapy. Participants in our sample are participants in the CCSS and, thus, likely represent a motivated and knowledgeable group. Accordingly, rates of diagnosis knowledge among childhood cancer survivors, in general, may well be lower.

Contrary to our a priori hypotheses, our analyses suggest that no consistent factors identify individuals with inadequate knowledge of their cancer diagnosis and therapy. Of note, no interaction was found between age at diagnosis and era of diagnosis. Also, individuals who attended a long-term follow-up clinic or received a medical summary, interventions receiving attention in the literature,[10,11] did not display greater understanding of their diagnosis and treatment. In fact many of the participants did not know whether they ever had received these interventions.

We observed particularly disturbing knowledge deficits in anthracycline exposure and site of radiation therapy, with lack of awareness among more than half of those who had received such treatments. These therapies are potentially associated with considerable toxic effects that warrant close monitoring, as reported in a consensus statement from the Cardiology Committee of the Children's Cancer Study Group.[12] Individuals who received anthracycline therapy are at risk for cardiomyopathy and early cardiac death. Depending on the cumulative anthracycline dose and the patient's symptoms, appropriate follow-up could range from a screening history and physical examination to sophisticated tests of cardiac function. Physicians evaluating these cardiac symptoms would provide better care to patients who had received anthracycline if patients were aware of that treatment. Similarly, radiation therapy is associated with long-term complications that require expert long-term medical follow-up, which depends on the site of radiation.[13] For example, radiation to the chest requires earlier and more frequent mammograms[3] in women.

The findings of this study must be understood within the context of some limitations. All diagnosis and treatment information used for comparison with self-report was obtained retrospectively by abstraction from the medical record. We attempted to limit errors by using quality-control assessments, which included re-abstraction of 5 subjects every 3 months at each CCSS institution for the first 15 months of the project. Furthermore, some of the data were determined at the time of the baseline survey several years ago. Individuals' income and educational levels may have changed. Similarly, it is possible that additional treatments have been administered since the medical data were abstracted. However, this is unlikely to affect the findings in our sample of underreporting and lack of crucial detail when describing past treatments already abstracted from the medical record. Finally, within some of the diagnostic categories with small sample size (eg, neuroblastoma), there was limited power to evaluate additional determinants of not knowing one's relevant medical history.

Compared with an earlier study[14] and 2 small, limited studies,[11,15] a greater proportion of participants in our study (93%) at least knew that they once had a cancer. Other investigators have reported rates in the range of 77% to 90%. The better rate of accurate diagnosis reporting in individuals treated in more recent years may reflect changing medical practice of being more candid with patients about their cancer diagnosis.[16] In our sample of participants in a long-standing study of childhood cancer survivors, we anticipated that a greater proportion of individuals would know the detailed name of their diagnosis.

Like Byrne et al,[14] we found highly accurate reporting of diagnosis by cases with Wilms tumor and Hodgkin disease, even after adjusting for age at diagnosis. Perhaps it is easier to remember an eponymic diagnosis. Also, individuals with Wilms tumor may receive additional contact through participation in the National Wilms Tumor Study group.

We found that survivors of CNS malignancies and neuroblastoma were significantly more likely to report their diagnosis and therapy inaccurately. Although not dramatic, we observed a general pattern of increased ORs within multivariate models containing factors significant in univariate analyses, such as age at diagnosis, educational attainment, and year of diagnosis. This suggests the potential of some confounding of the associations observed for these 2 diagnostic groups.

Respondents with a history of CNS cancer were the least informed with only 84% knowing that they had a cancer (ie, naming a noncancer or tumor condition or responding that they did not know). This finding may not be surprising given the well-documented cognitive and psychosocial sequelae of treatment for CNS malignancies.[8] Interestingly, our data suggest that radiation to the head or neck region was not a separate risk factor for reduced knowledge in this population. Many of these survivors of CNS cancer may have been primarily managed by neurosurgeons with, potentially, less opportunity for long-term follow-up. Because of neuropsychological effects, this subgroup of individuals who had cancer as children may require educational interventions tailored to their special needs.

Adding probing history questions about cancer diagnosis to prompt the survivor's memory only improved the quality of the response slightly. Almost one third of participants in our study gave nondetailed or inaccurate responses, which would render risk for future disease complications difficult. Subtypes of leukemia, lymphoma, and

brain tumors are associated with differing long-term issues because of the different therapies administered.[8]

Radiation therapy (especially to the head or neck and spine) was falsely reported by 10% of respondents. We speculate that they confused diagnostic radiological procedures with therapeutic ones. Perhaps understanding this difference led to less overreporting by better-educated respondents. Overreporting of these therapies could be deleterious. Unnecessary monitoring for endocrine, cognitive, pulmonary, and cardiac complications would be expensive. Furthermore, the patients could experience undue anxiety and stress.

Survivors of Hodgkin disease generally were not as well-informed about their history of splenectomy as they were about other aspects of their cancer history. One third of individuals who had a splenectomy did not report it when asked to list their surgeries. Determining which patients had undergone a splenectomy is important so that appropriate immunization against encapsulated organisms and timely antibiotics can be administered.[17]

Like Hudson et al,[18] we found that a minority of individuals were anxious about the potential deleterious late effects related to their therapy. In our study, only one third of respondents believed that treatment for a previous childhood malignancy could cause serious health problems as they grew older. Among those with awareness of potential late effects, there was no greater level of knowledge about diagnosis or therapy. These findings suggest that this population should be the target of future educational interventions.

A rapidly expanding literature continues to identify medical complications that affect adult survivors of childhood cancer. Diligent screening and appropriate treatment can reduce the morbidity and mortality of these late effects. Fortunately, many individuals do know some details about their diagnosis and treatment. However, medical care providers must recognize that much of a patient's history, even with probing questions, can not be trusted to guide medical management. Medical records should be obtained from the treating institution prior to formulating long-term follow-up care, whenever possible. Survivors should be better educated about their medical history to be motivated to pursue appropriate follow-up. Treatment summaries must be given to patients in a form that is accessible, even many years later. Perhaps, with the advent of the information age, a secure electronic record may be a feasible option. Finally, further studies are needed to determine how long-term follow-up clinics can better educate patients.

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