

The Met and Unmet Health Care Needs of Adult Survivors of Childhood Central Nervous System Tumors

A Double-Informant, Population-Based Study

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BACKGROUND: The purpose of the current study was to examine the persistent health care needs (HCNs) of adult survivors of childhood central nervous system tumors. **METHODS:** In this population-based study, 526 of 679 eligible survivors and 550 parents provided data. Survivors' HCNs were assessed using a questionnaire covering 4 domains: Medical Care, care coordination and communication (Care Coordination), Illness Education, and Psychosocial Services. Needs were categorized as no need, met need, and unmet need. Outcomes were analyzed specifically in relation to survivors' functional late effects as assessed using the Health Utilities Index Mark 2/3. **RESULTS:** Approximately 40% of survivors experienced their HCNs as exceeding the supposed general population average, and 41% had a current HCN that was unmet. The most common unmet need concerned the Psychosocial Services domain (reported by 40%), followed by a lack of Illness Education (35%), Care Coordination (22%), and Medical Care (15%). Survivors experiencing functional late effects had greater HCNs, and a greater percentage of unmet needs. Agreement between survivor-reported and parent proxy-reported HCNs was satisfactory, whereas agreement for survivors' unmet HCNs ranged from poor to satisfactory. **CONCLUSIONS:** Findings based on reliable double-informant data demonstrated that a considerable percentage of adult survivors report unmet HCNs, with female sex, younger age at diagnosis, and indications of disability and poor health status comprising significant risk factors. Issues critical for improved, comprehensive, long-term follow-up care were identified. Addressing these issues adequately in clinical follow-up extending into adulthood would likely improve the quality of comprehensive care for this patient group. *Cancer* 2011;117:4294-303. © 2011 American Cancer Society

KEYWORDS: childhood cancer, central nervous system tumors, adult survivors, health care needs, long-term follow-up, aftercare.

Advances in childhood cancer diagnostics and treatment have resulted in a growing population of long-term survivors. The 5-year survival rate after treatment according to recent protocols is > 75%.¹ Survivors of childhood cancer require follow-up health care because of their relatively heightened susceptibility to late effects, some of which may become apparent several years after the end of treatment.²⁻⁵ Indeed, studies of health care use in survivors have indicated significantly greater needs for health care services compared with the general population.⁵ In a long-term perspective, additional risks may follow with adulthood and aging, risks that require intensified medical attention.

Providing appropriate health care for childhood cancer survivors is a recognized major emerging challenge in pediatric oncology.⁶ The challenge is related to the heterogeneity of the patient group, and variations in treatment. The goals of long-term follow-up include monitoring late effects, and providing education about illness and psychosocial services.⁷ An understanding of survivors' unique needs and how they may vary with diagnosis, late effects, age, and sex is fundamental in achieving the goals of surveillance and long-term follow-up care.

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A majority of studies on health care and follow-up needs have addressed childhood cancer survivors as a uniform group,^{5,8-11} without distinguishing between diagnostic subgroups, despite an established factual variation in aftercare and treatment-related symptoms depending on diagnosis.¹² Approximately 67% of childhood cancer survivors have been found to experience at least 1 late complication, with as many as 25% to 40% of these survivors presenting with severe or even life-threatening chronic conditions.³ Compared with the general population and with survivors of other childhood cancers, survivors of central nervous system (CNS) tumors comprise a high-risk population with regard to tumor- and treatment-related sequelae,^{2,3,13} with socioeconomic consequences and substantial dependence on social services in adult life.^{14,15} The current 10-year survival rate for childhood CNS tumors in Sweden is 72%,¹⁶ meaning that the majority of patients will become long-term survivors. Although they are at risk for multiple sequelae, to our knowledge little is known regarding how this fact may result in a heightened need for extended health care in later life. To our knowledge, the perceived health care needs (HCNs) of adult survivors of childhood CNS tumors have not been specifically addressed in prior large-scale, population-based studies.

The aim of the current study was to perform a comprehensive investigation of the HCNs of adult survivors of childhood CNS tumors. The specific aims were to identify unmet HCNs and to examine HCNs in relation to status of late effects specifically, and other potentially modifying factors (sex, treatment center, unmet informational needs, use of social subsidies, time since diagnosis, age at diagnosis, age at assessment, and era of treatment).

Survivorship studies have mostly relied on patient-reported information only. However, reliance on self-report data can result in biased outcomes.^{17,18} In the current study, data were collected from both survivors and family caregivers to increase reliability and arrive at a more comprehensive evaluation.¹⁹

MATERIALS AND METHODS

Participants and Procedures

This population-based cohort study involved all 6 childhood cancer centers in Sweden, and included survivors and their parents. Participants were identified through the Swedish Childhood Cancer Registry (SCCR), which contains information concerning primary cancer diagnoses

classified according to the third edition of the International Classification of Childhood Cancer.²⁰ Survivors who met the following criteria were eligible: a primary CNS tumor diagnosis received between 1982 and 2001 before the patient's 19th birthday, > 5 years elapsed from the time of diagnosis, and patient age \geq 18 years at the time of assessment. Of the 1535 children diagnosed with a CNS tumor between 1982 and 2001, 708 patients were eligible. Of these, 11 had died since the last update of the SCCR, and 18 survivors and 22 parents could not be reached. Among the remaining 679 survivors, 531 returned questionnaires. Responding ($n = 531$) and nonresponding survivors ($n = 148$) were similar with regard to age at invitation, time since diagnosis, sex, and diagnosis, but differed with regard to age at diagnosis ($t_{216} = 2.30$; $P = .02$). Nonresponders were younger at the time of diagnosis (mean age, 9.52 years; standard deviation [SD], 4.97 years) compared with responding survivors (mean age, 10.56 years; SD, 4.43 years). Five incomplete survivor questionnaires were excluded, resulting in a final study sample of 526 survivors (77.5%). Parent proxy ratings of survivors' HCNs were obtained from 556 returned questionnaires. Responding ($n = 556$) and nonresponding ($n = 119$) parents were similar with regard to the child's sex, diagnosis, age at diagnosis, and time since diagnosis, and child and parent ages at follow-up. Six parent questionnaires were incomplete, resulting in a final sample of 550 parents (81.5%). Table 1 presents characteristics of the study group. Data collection took place between June 2006 and March 2007, after participants provided informed consent. The study was approved by the Regional Research Ethics Committee.

Assessments

Sociodemographic data were collected using a self-report form encompassing academic attainment, employment, and use of social insurance or governmental subsidies. Medical information was obtained from the SCCR.

HCNs

Survivors and parents completed an 11-item questionnaire covering 4 domains of HCNs: Medical Care, care coordination and communication (Care Coordination), Illness Education, and psychosocial and social counseling (Psychosocial Services) (Table 2). The questionnaire, originally developed by Stein and Jessop,²¹ has since been further developed for the study of satisfaction with health care.²² An opening question preceding the questionnaire asked whether the survivor's need of health care was

Table 1. Study Group Characteristics

Survivor Characteristics	Total (n=526)	Males (n=272)	Females (n=254)	P^a
Age at assessment, y				
Mean/median (SD)	26.3/25.6 (5.0)	26.3/25.4 (5.1)	26.3/26.1 (4.8)	.950
No. (%)				.524
<25	235 (44.7)	124 (45.6)	111 (43.7)	
25-30	194 (36.9)	98 (36.0)	96 (37.8)	
31-36	79 (15.0)	38 (14.0)	41 (16.1)	
>36	18 (3.4)	12 (4.4)	6 (2.4)	
Age at diagnosis, y				
Mean/median (SD)	10.6/11.3 (4.4)	10.9/11.8 (4.4)	10.2/11.0 (4.5)	.080
No. (%)				.590
<5	72 (13.7)	34 (12.5)	38 (15.0)	
5-10	149 (28.3)	76 (27.9)	73 (28.7)	
10-15	213 (40.5)	109 (40.1)	104 (40.9)	
>15	92 (17.5)	53 (19.5)	39 (15.4)	
Time since diagnosis, y				
Mean/median (SD)	15.8/16.2 (5.2)	15.4/15.7 (5.1)	16.2/16.7 (5.3)	.088
No. (%)				.531
<10	90 (17.1)	50 (18.4)	40 (15.7)	
10-15	130 (24.7)	72 (26.5)	58 (22.8)	
15-20	180 (34.2)	87 (32.0)	93 (36.6)	
>20	126 (24.0)	63 (23.2)	63 (24.8)	
Diagnosis, no. (%)				.087
Ependymoma	44 (8.4)	18 (6.6)	26 (10.2)	
Astrocytoma/glioma	279 (53.0)	136 (50.0)	143 (56.3)	
PNET/medulloblastoma	68 (12.9)	41 (15.1)	27 (10.6)	
Other	135 (25.7)	77 (28.3)	58 (22.8)	
Education, no. (%)				.025
< High school graduate	82 (15.6)	40 (14.7)	42 (16.5)	
High school graduate	182 (34.6)	109 (40.1)	73 (28.7)	
College graduate/postgraduate education	261 (49.6)	123 (45.2)	138 (54.3)	
Missing	1 (0.2)	0	1 (0.4)	
Parent Responder, no. (%)				
Mother	406 (73.8)			
Father	30 (5.5)			
Both	112 (20.4)			
Missing	2 (0.4)			

SD indicates standard deviation; PNET, primitive neuroectodermal tumor.

^aP values for comparisons by sex.

judged to be greater than the average person's in the community because of one's past illness and treatment.

Respondents indicated on a 4-point scale whether they/their child: 1) received the health service; 2) received it partly; 3) did not receive it, and had no need for it; or 4) did not receive it, but had a need for it. HCNs were categorized as met need (response categories 1 and 2), no need (response category 3), and unmet need (response category 4). A HCN was considered to be present for those who

responded positively regarding response alternatives 1, 2, or 4. If the respondent answered "do not receive the service but need it" to at least 1 question within a domain, an unmet HCN in that domain was recorded. The same approach was used when classifying existing HCNs within a specific domain. The Psychosocial Services domain comprises a greater number of items than the other domains (Table 2). Therefore, a need verified in response to at least 2 questions was required for a HCN to be

Table 2. Proportion of Survivors with No HCN, A Present HCN, and an Unmet HCN

Domain	Items	Valid Responders	No Need No. (%)	Having Need No. (%)	Unmet Need No. (%)
Medical Care	1. Do you have a physician who regularly takes care of your health needs?	520	191 (36.7)	329 (63.3)	42 (12.8)
	2. Does anyone supervise your growth and development?	522	245 (46.9)	277 (53.1)	35 (12.6)
Care Coordination	3. Does your physician make arrangements if you need to see a specialist?	508	155 (30.5)	353 (69.5)	43 (12.2)
	4. Does your physician communicate with school, work, or other therapists?	516	371 (71.9)	145 (28.1)	62 (42.8)
Illness Education	5. Has your condition, including possible physical and/or developmental delay or psychological disability, been sufficiently explained?	524	157 (30.0)	367 (70.0)	96 (26.2)
	6. Does anyone counsel you about special appliances?	521	314 (60.3)	207 (39.7)	86 (41.5)
Psychosocial Services	7. Does anyone provide you with general advice about possible educational or behavioral problems?	521	346 (66.4)	175 (33.6)	97 (55.4)
	8. Can you talk to someone who understands the special problems of being a person with a potential illness or disability?	525	239 (45.5)	286 (54.5)	71 (24.8)
	9. Were you provided with information about patient groups or self-help organizations?	518	334 (64.5)	184 (35.5)	89 (48.4)
	10. Were you provided with information about social services?	523	313 (59.8)	210 (40.2)	86 (41.0)
	11. Did you receive information about the Swedish Act concerning Support and Service for Persons with Certain Functional Impairments?	523	372 (71.1)	151 (28.9)	53 (35.1)

HCN indicates health care need.

recorded for Psychosocial Services, and at least 2 unmet needs had to be verified for an unmet need to be recorded.

Functional Late Effects

We used the 15-item Health Utilities Index Mark 2/3 (HUI2/3) for assessing functional late effects. The HUI2/3 has proven to be applicable across age groups and in various clinical and nonclinical populations.^{23,24} It allows for the establishment of an overall health status score, and this was used in the current study. The measure is based on a theoretical framework that describes individuals' health on a scale, in which death is indicated by 0.00 and perfect health by 1.00. Health status scores for states characterized by mild disability fall into the range of 0.89 to 0.99 and states with moderate disability into the range of 0.70 to 0.88, whereas states with severe disability have scores < 0.70.^{23,24}

Information Satisfaction

Survivors' satisfaction with the extent of information provided during treatment and follow-up was assessed using a summary item from a questionnaire adapted for this

study, and based on issues covered in the European Organization for Research and Treatment of Cancer (EORTC) QLQ-INFO26.²⁵ If survivors reported unmet information needs, they were asked to specify the kind of information they felt a need for.

Statistical Analyses

We provided descriptive summary statistics for the demographic characteristics of survivors. Reported HCNs are presented using valid percentage, ie, based on those who actually responded to the specific question. Frequency statistics for unmet HCNs were based on survivors and parents who initially indicated a need for the specific service or domain.

Differences in demographic and cancer-related variables between responders and nonresponders were analyzed using the Student *t* test for independent groups and chi-square tests. Chi-square tests were conducted to analyze associations between HCNs and late effects status, information satisfaction, and sociodemographic variables. The categorization of variables shown in Tables 1 and 3 was maintained in the analyses.

Table 3. Sociodemographic and Late Effects Status Outcomes

Outcome	Total (n=526)	Males (n=272)	Females (n=254)	P ^a
Employment status, no. (%)				
Employed/studying	383 (72.8)	208 (76.5)	175 (68.9)	.080
Unemployed	136 (25.9)	62 (22.8)	74 (29.1)	
Missing	7 (1.3)	2 (0.7)	5 (2.0)	
Use of social insurance/government subsidies, no. (%)				
Yes	174 (33.1)	77 (28.3)	97 (38.2)	.043
No	342 (65.0)	190 (69.9)	152 (59.8)	
Do not know	3 (0.6)	1 (0.4)	2 (0.8)	
Missing	7 (1.3)	4 (1.5)	3 (1.2)	
Status of late effects, no. (%)				
Perfect health	85 (16.2)	52 (19.1)	33 (13.0)	.003
Mild disability	159 (30.2)	95 (34.9)	64 (25.2)	
Moderate disability	141 (26.8)	69 (25.4)	72 (28.4)	
Severe disability	129 (24.5)	52 (19.1)	77 (30.3)	
Missing	12 (2.3)	4 (1.5)	8 (3.1)	

^aP values for comparisons by sex.

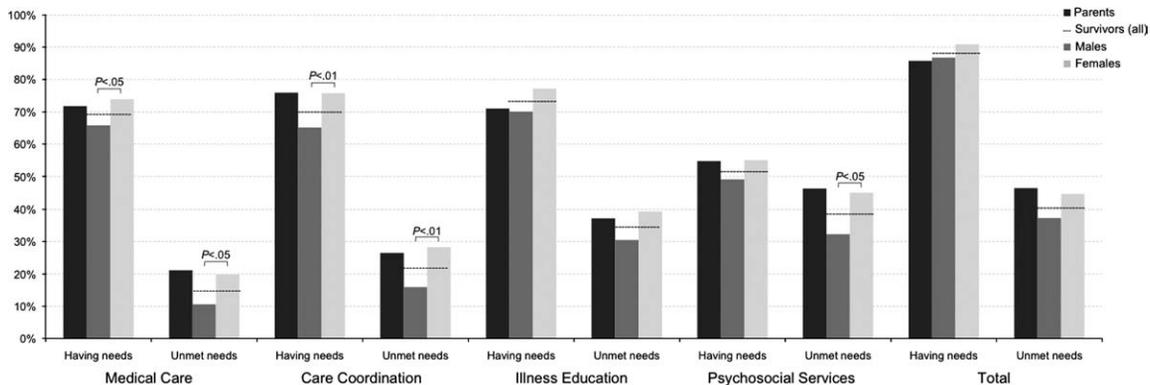


Figure 1. Percentage of survivors with health care needs (HCNs) and unmet HCNs is shown by sex, based on survivor-reported (n = 526) and parent proxy-reported (n = 550) data. The percentage of unmet needs is based on survivors and parents who initially indicated a need in the specific domain. Dotted line indicates survivor-reported outcomes (males and females); P value brackets, localization of sex differences.

Agreement between survivor reports and parent reports was evaluated using kappa statistics, and by presenting the percentage agreement. Kappa values between 0.41 and 0.75 represent moderate to good agreement, and a kappa statistic of ≥ 0.75 represents substantial to excellent agreement.²⁶

Two-tailed testing and an alpha level of .05 were applied for statistical significance. Analyses were performed using the SPSS statistical package for Windows (version 17.0; SPSS Inc, Chicago, Ill).

RESULTS

Table 3 presents sociodemographic and late effects status outcomes. Functional late effects were significantly more prominent among female survivors compared with male

survivors (Table 3). Health and functional status outcomes have been presented in more detail elsewhere.²

HCNs

Overall, in response to the opening question preceding the 11-item scale, 39% of survivors (n = 192) reported that due to their illness and treatment, their need for health care exceeded the supposed average of the general population.

Approximately 89% of survivors (n = 467) indicated some kind of current HCN by expressing either that the health service had been received when needed (fully/partly received) or by expressing an unmet need (Fig. 1). The greatest survivor-reported needs were found for the Illness Education domain (n = 386), followed by Care

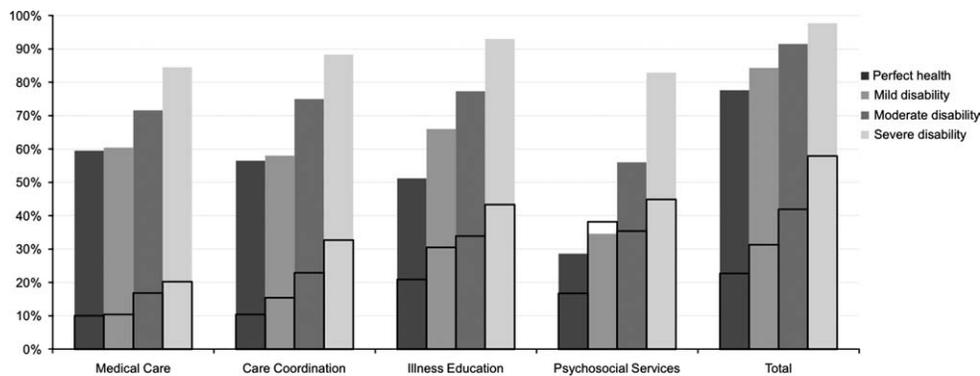


Figure 2. Percentage of survivors with unmet health care needs (HCNs) (indicated by transparent overlay bars) are shown by late effects status. The colored background bars and the foreground transparent bars refer to different subgroups; background bars indicate the percentage of survivors with a HCN, whereas foreground bars indicate the percentage of survivors with unmet HCNs.

Coordination ($n = 367$), Medical Care ($n = 366$), and Psychosocial Services ($n = 273$). The most frequently reported specific needs concerned information regarding their illness-related condition, including possible late effects, followed by the need for access to a physician for referrals to a specialist when necessary (Table 2). The quest for information concerning illness and treatment, and late effects in particular, was verified by the answers to the open-ended question regarding where information was experienced as being insufficiently provided.

Overall, of the survivors who indicated that they had a HCN, 41% ($n = 191$) reported that they also had an unmet HCN (Fig. 1). Most frequently, unmet needs were found for the Psychosocial Services ($n = 106$; 39% of those having a need) and Illness Education ($n = 135$; 35% of those having a need) domains. The most frequent specific unmet HCNs concerned the lack of opportunity for counseling regarding educational or behavioral problems, and insufficient information regarding survivor meeting groups or self-help organizations (Table 2). Among the 232 survivors who specified the issues for which they had additional information needs beyond what was satisfied by the provided information, 42% mentioned late effects, 21% cited their illness in general, and 13% mentioned treatment-related issues. Furthermore, insufficiently met information needs were frequently reported regarding the causes of illness (12%) and the possibilities for psychological (10%) and rehabilitation (9%) services.

HCNs and Late Effects Status

Survivors ($n = 192$) who considered their need for health care to exceed the population average because of their

past CNS disease experienced more functional late effects compared with survivors with no such pronounced HCNs ($n = 296$) (chi-square test, 83.36; $P < .001$).

The percentage of survivors who reported having HCNs in adult life differed according to their current late effects status (Fig. 2). Survivors with severe disability had the most prominent needs, followed by survivors with moderate, mild, and no disability (total: chi-square, 24.57; $P < .001$).

The percentage of survivors with perceived unmet HCNs differed by survivors' late effects status with regard to the Care Coordination (chi-square, 13.51; $P = .004$) and Illness Education (chi-square, 8.42; $P = .038$) domains, but not with regard to the Medical Care ($P = .167$) and Psychosocial Services ($P = .072$) domains (Fig. 2). In total, 58% of survivors ($n = 73$) with severe disability had an unmet need compared with 42% of survivors ($n = 54$) with moderate disability, 31% of survivors ($n = 42$) with mild disability, and 23% of survivors ($n = 15$) with no disability (chi-square, 29.32; $P < .001$).

Other Modifying Factors

The percentage of survivors who reported that their need for health care exceeded the general population average ($n = 192$) was greater among females than males (46% vs 33%; chi-square, 9.23 [$P = .002$]).

Furthermore, female survivors reported more HCNs with regard to the Medical Care and Care Coordination domains (Fig. 1). Female survivors also reported more unmet HCNs regarding the Medical Care, Care Coordination, and Psychosocial Services domains (Fig. 1).

Item-wise analysis of the observed sex differences indicated that female survivors reported greater specific HCNs as covered by 5 single items (items 1, 3, 4, 5, and 6 in Table 2): physician care (70% of females and 58% of males indicated a need; chi-square, 7.93 [$P = .005$]), specialist referrals (75% of females and 64% of males indicated a need; chi-square, 7.03 [$P = .008$]), physician communication (34% of females and 23% of males indicated a need; chi-square, 5.46 [$P = .005$]), condition sufficiently explained (74% of females and 66% of males indicated a need; chi-square, 4.49 [$P = .034$]), and counseling about special appliances (44% of females and 36% of males indicated a need; chi-square, 5.46 [$P = .047$]).

In terms of unmet HCNs, the percentage of male and female survivors differed with regard to 4 specific HCNs (items 1, 3, 4, and 9 in Table 2): physician care (18% of females and 7% of males indicated a need; chi-square, 8.70 [$P = .003$]), specialist referrals (17% of females and 7% of males indicated a need; chi-square, 7.82 [$P = .005$]), physician communication (50% of females and 33% of males indicated a need; chi-square, 4.28 [$P = .039$]), and information regarding patient groups/self-help organizations (59% of females and 36% of males indicated a need; chi-square, 9.82 [$P = .002$]).

The survivors who had information needs beyond what was satisfied ($n = 261$) also had greater unmet HCNs compared with survivors with satisfied information needs ($n = 232$) (total: 57% vs 23%; chi-square, 51.97 [$P < .001$]).

In addition, survivors who received social insurance or governmental subsidies ($n = 174$) had greater HCNs compared with survivors not receiving economic aid ($n = 342$) (total: 94% vs 86%; chi-square, 8.38 [$P = .004$]). Furthermore, those survivors receiving such subsidies had greater unmet HCNs compared with survivors not receiving economic aid (total: 53% vs 35%; chi-square, 4.99 [$P < .001$]).

A younger age at diagnosis was found to be related to having more HCNs for the Psychosocial Services domain (chi-square, 12.72; $P = .005$) and having unmet needs regarding the Care Coordination domain (chi-square, 12.42; $P = .006$). The percentage of survivors with HCNs or unmet needs did not differ significantly with regard to treatment centers, era of treatment, time since diagnosis, or age at assessment.

Informant Agreement

Parents' and survivors' reports demonstrated a similar pattern regarding the extent to which HCNs and unmet

Table 4. Survivor and Parent Proxy Agreement for Participants With Both Parent- and Survivor-Reported Data Available ($n=472$)

Domain		Percentage Agreement	Kappa
Medical Care	Having needs	84.5	0.63
	Unmet needs ^a	85.5	0.46
Care Coordination	Having needs	80.9	0.52
	Unmet needs	77.2	0.36
Illness Education	Having needs	74.8	0.37
	Unmet needs	68.8	0.33
Psychosocial Services	Having needs	78.5	0.57
	Unmet needs	63.4	0.26
Total (all domains)	Having needs	88.3	0.50
	Unmet needs	70.1	0.39

HCNs indicates health care needs.

^aAnalyses for unmet needs based on sets of data in which both informants initially indicated a need (Medical Care: $n=297$; Care Coordination: $n=294$; Illness Education: $n=279$; Psychosocial Services: $n=194$; and Total: $n=381$).

HCNs appeared over the domains (Fig. 1). At the entire-group level (survivors and parents, unpaired), the only significant differences between survivors' self-reported data ($n = 526$) and parent-reported data ($n = 550$) were found for the extent of HCNs with regard to the Care Coordination domain (chi-square, 4.21; $P = .040$), and for unmet needs in the Medical Care domain (chi-square, 4.22; $P = .040$). Here, parents reported greater HCNs and more unmet needs than did survivors themselves.

Survivor and parent proxy agreements for participants for which both parent- and survivor-reported data were available ($n = 472$ pairs) are presented in Table 4. Agreement was higher for extent of HCNs compared with reported unmet needs. Kappa statistics for survivors' needs were satisfactory, ranging from 0.37 to 0.63. Kappa statistics for unmet HCNs varied between 0.26 and 0.46, indicating poor to satisfactory agreement.

DISCUSSION

To our knowledge, the current study is the first to systematically evaluate the perceived needs of health care in a Swedish nationwide cohort of adult survivors of childhood CNS tumors. Approximately 40% of the survivors reported that they, compared with the general population, had heightened HCNs because of their past illness and treatment. Among the approximately 90% of survivors who indicated having some kind of HCN in adult life, a substantial percentage (approximately 40%) found their needs to be unmet. Survivors with persistent health-related late effects had greater HCNs as well as more unmet HCNs.

A considerable percentage of the adult survivors stated that, because of their childhood CNS tumor and the cancer treatment, they had greater HCNs than assumed for the general population. This finding confirms the necessity of extended surveillance and medical aftercare that continues into adulthood for these patients. Although use of health care services was not investigated, the findings of the current study not only illustrate the presence of heightened needs, but also of a most likely heightened use of health care services by this population. In the current study, HCNs were investigated using a multidimensional questionnaire covering central health care issues for which the services of the common medical care system are responsible. The most frequent needs concerned information and knowledge about one's illness. This finding parallels those from prior studies demonstrating the importance survivors attach to having individualized information regarding long-term health issues.^{4,27,28} The findings of the current study indicate that there also is a prominent need for such information among adult survivors of childhood CNS tumors. Illness information has been acknowledged both by patients and professionals as a vital part of regular care and surveillance.²⁷ The results of the current study indicate that more than one third of survivors experienced unmet HCNs in the Illness Education domain. This was particularly prominent among survivors with severe disabilities. These findings are of special concern, because inadequate information regarding cancer treatment and treatment-related health risks have been found to be associated with poor compliance with regular surveillance.^{5,8,11} The chances of obtaining adequate long-term follow-up have in prior studies been found to decrease with age,^{5,11} whereas the incidence of late effects may increase with time.³ Although many survivors may have fair knowledge concerning their past diagnosis and treatment, the current study findings demonstrate that too few have been provided with sufficient information regarding the long-term risks of cancer therapy. This indicates a need for extended and updated information as part of the follow-up care of survivors of childhood CNS tumors. Some type of a "survivor pass," a document issued from the treating institution containing individualized information regarding their illness, including possible late effects, could be a way of facilitating the proper management of persistent and late-occurring HCNs once standard aftercare has been completed.

Patients of the studied cohort were followed at pediatric oncology centers at least once a year during the first

5 years after diagnosis (earliest era of treatment) or until their 18th birthday (later era of treatment). Over time, and particularly from approximately the year 2000 onward, when centers started following the national guidelines for medical and psychological follow-up, aftercare has become more regular, and follow-up visits more frequent. In Sweden, when patients have been discharged from pediatric medical care, long-term care is problem oriented, based on the survivor's condition. Many patients without overt HCNs are not offered further follow-up, or have to rely on their own initiative for physician appointments. A substantial percentage of the adult survivors in the current study experienced unmet HCNs as a result of this organization of long-term follow-up care. The most frequent unmet needs were those concerning the domain of Psychosocial Services. This has also been verified in prior studies of adjacent study groups.⁹ The population-based outcomes permit a reliable identification of areas in need of improvement. A prominent unmet psychosocial need concerned insufficient information regarding survivor groups and patient self-help organizations. Although such activities may be more available for younger survivors, the findings of the current study indicate that opportunities for meeting others who share a history of cancer in later life are insufficient.

Being female and suffering from compromised functional ability were found to be associated not only with having greater HCNs, but also with having more unmet HCNs. The greater unmet long-term HCNs noted in females compared with males parallel prior findings regarding adult cancer patients and survivors of other childhood malignancies.^{12,29} The observed sex differences could, in part at least, be explained by a greater incidence of late effects reported by the female survivors. However, because the sex-related differences may have additional or other explanations not illuminated by the current study data, we view these outcomes as an important target for further study.

Current guidelines for long-term follow-up pay respect to individual variations in surveillance needs. In keeping with such a needs-oriented strategy, the established relation between functional disabilities and increased HCNs suggests that a health status screening such as that performed by us functions as a means of customizing long-term aftercare, including psychosocial services and education about illness. The finding that survivors with identified late effects had greater HCNs might be expected. However, the association between impaired health and unmet HCNs was less anticipated

and has obvious clinical implications. This finding clearly indicates that follow-up care should be intensified and extended for the group of patients experiencing compromised health in adulthood.

The major strengths of the current study include the population-based approach, a high response rate, and the data based on dual responders. The use of double informants contributes to an understanding of how far survivors may under-report or “deny” problems, and adds reliability to the evaluation. Agreement between survivor-reported and parent proxy-reported HCNs was satisfactory, whereas poorer agreement was established for reported unmet HCNs. Parents tended to report greater unmet needs than did survivors themselves, an observation that demonstrates the value of using double informants to avoid potential bias due to under-reporting of difficulties.¹⁷ In addition, because some survivors are unable to formulate their HCNs due to disability, the proxy ratings importantly allow for a more complete evaluation.¹⁹

Certain limitations should be considered when interpreting the findings of the current study. First, the unmet HCNs were self-reported, and we did not use other sources regarding survivors’ access to the studied health care services. At the same time, the value of investigating the self-reported experience of health care is obvious, because the extent of HCNs and unmet needs could hardly be fully identified without addressing survivors’ own experiences. Second, we had no control group for information regarding the HCNs of the general population. The questionnaire used is an illness-specific measure. As such, it is not appropriate for use in nonclinical groups. Although adult survivors of childhood CNS tumors have indeed been found to demonstrate a heightened need for aid and support compared with the general population in Sweden,¹⁴ the results of the current study add information regarding the unique needs of these patients. Third, the current study findings regarding unmet HCNs may not be in all parts fully generalizable to countries in which follow-up care is different. Finally, although the questionnaire was constructed to encompass needs associated with medical and psychosocial follow-up, we cannot be sure that certain HCNs remained undetected.

In conclusion, addressing the HCNs of adult survivors of childhood CNS tumors becomes increasingly important as the population of long-term survivors increases and the long-term adverse effects of former and novel treatments are unknown. The results of the current study identify and describe both the self-reported HCNs and

unmet HCNs of adult survivors of childhood CNS tumors, a high-risk population for whom the unique HCNs have not been addressed previously. The outcomes provide evidence-based guidance for a more effective and individualized follow-up extending into adulthood, with findings that in many aspects may well apply to survivors of non-CNS tumors as well. The results of the current study indicate that certain areas of follow-up care can be improved at an affordable cost to better satisfy the needs of adult survivors of childhood CNS tumors, even when resources are limited. Identified unmet HCNs indicate that the domains of Illness Education and Psychosocial Services are primary targets for improvement. Furthermore, the results of the current study verify the importance of adequate psychological services being regularly integrated into ordinary long-term follow-up. These findings should be useful for all health care providers when developing empirically based strategies for long-term survivor care. We also believe in the general applicability of the importance survivors placed on comprehensive follow-up, including both medical and psychosocial care, with the latter currently at risk of being insufficiently provided.

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