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Persistent impact of illness on families of adult survivors of childhood central nervous system tumors: a population-based cohort study

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Abstract

***Objective*: This study aims to determine the long-term impact on families of adult survivors of childhood central nervous system tumors. Illness-related family consequences were studied in relation to modifying determinants.**

***Methods*: In a population-based cohort of parents of 697 survivors 18 years and older, 551 parents provided data. The impact of cancer on the families was evaluated in four domains using the Impact on Family Scale (economic situation, personal burden, social life, sibling impact). The results were analyzed in relation to survivors' health assessed using the Health Utilities Index™, parent satisfaction with information about illness and treatment, and perceived health-care needs of their child.**

***Results*: Despite an established mild-to-moderate impact on the group level, outcomes provided evidence of substantial cancer-related family consequences even once the child had reached adulthood. About one fifth of parents reported psychological and financial difficulties exceeding the cutoff limit for a significant impact still ≥5 years after diagnosis. A stronger total family impact was associated with poorer health of survivors ($F[3,302] = 56.65$, $p < 0.001$), and unmet informational - ($F[3,231] = 14.06$, $p < 0.001$) and health-care needs ($t_{218} = 5.31$, $p < 0.001$). The impact was unrelated to survivors' age at follow-up and time since diagnosis.**

***Conclusions*: Adverse cancer-related consequences affect a considerable portion of families of childhood survivors of central nervous system tumor, even after reaching adulthood. The impact is aggravated by lasting sequelae and perceived shortcomings of long-term follow-up, factors that partly are avoidable. Improved clinical follow-up should particularly address illness information and long-term health-care needs to reduce the impact on families of survivors suffering from chronic health conditions.**

Keywords: oncology; childhood cancer; CNS tumors; family impact; parents; adult survivors

Introduction

Childhood cancer and its treatment are potential sources of distress and result in an altered life situation for all family members. Although soon after diagnosis and the period of active treatment are associated with greatest parental distress [1–3], many parents continue to experience cancer-related stressors even years after treatment completion that may result in ongoing concerns [4–6]. Because of the lack of large-scale studies, the extent to which such stressors and other factors influence the burden on the family has yet to be uncovered. The determinants of family consequences are not fully known, although factors related to illness complication, the child's social functioning, distance to specialized treatment centers, and parental psychosocial factors have all been pointed out as potentially influential [7–14]. Although chronic conditions that affect neurological functioning can be particularly challenging [15,16], parents may worry about relapse and their child's development even in the absence of sequelae [17,18]. Continuing uncertainty regarding late effects and lifetime expectancy typically remain parental stressors after completion of cancer treatment [19–21]. Along with such ongoing stressors, the severity of the illness and a need of continued follow-up result in that many families need to adapt to a life situation characterized by heightened caregiving demands [22–25]. Continued involvement in monitoring of survivors' health and follow-up constitutes a significant part of such situation-specific demands parents face [16,24,26]. The measure used for assessing family impact in this study provides knowledge about the changes families of adult survivors need to do to meet those demands. The nature and severity of family consequences in areas that are specifically relevant in the childhood cancer situation also informs about these families' quality of life in a broader sense. Identifying areas of adverse family impact is a precondition for the implementation of prophylactic interventions that facilitate families' adaption to the requirements of the illness situation and the post-treatment life.

Advances in diagnostics and treatments have resulted in a growing number of long-term pediatric survivors of central nervous system (CNS) tumors [27,28]. Compared with the general population and other childhood cancers, patients with CNS tumor constitute a high-risk population regarding late effects, some persisting into adulthood or appearing late [29–34]. This heightened risk of sequelae is particularly threatening for parents [11,21,35].

Prior studies of the impact of childhood cancer on the family have typically involved limited samples sizes and parents of young, non-adult patients [36–40]. Although parents of newly diagnosed patients, adolescent, and young adult survivors have been addressed [4,41], with few exceptions [18,42,43], families of older survivors have remained understudied. The responsibilities of parents of children suffering from irreversible morbidity or even a mild disability do not end with the child's transition to adulthood. The extent to which the increased vulnerability for late effects of CNS tumor patients has consequences for the family, and the unique continuing conditions of parents of adult survivors have, to our knowledge, not previously been investigated. Parents of young brain-tumor survivors have indeed been found to display post-traumatic stress, distress, and heightened concern about relapse, their child's health, and ability to take care of himself or herself [16,19–21,35,44,45]. Whether such findings are related to the adverse impact on families of adult survivors has not been studied, resulting in gaps in the knowledge necessary for the planning of appropriate long-term follow-up, assistance, and support interventions.

Whereas most previous studies have particularly focused on psychosocial consequences for parents of childhood CNS tumor survivors, this study investigated and quantified the adverse impact on families. We, thus, addressed the persistent burden of continued involvement in health monitoring and care taking in terms of impacts on the family. Impact was conceptualized as the consequences of the child's illness on the family system [46], specifically in terms of changes in the family with regard to social life, interaction with significant others, subjective distress or strain, time for other family members (siblings), and financial status. The specific aims were to use standardized methods to assess the persistent impact in these domains. In addition, we wanted to identify potential determinants of the impact including a history of relapse, survivors' sex, age of survivors and parents, age at diagnosis, time elapsed since diagnosis, survivors' health status, perceived unmet health-care needs, satisfaction with illness-related information, and the distance to a specialized treatment/follow-up center.

Methods

Study population and procedure

This population-based cohort study covered all eligible parents of CNS tumor survivors treated at any of six childhood cancer centers in Sweden. It was part of a larger investigation of long-term outcomes of childhood CNS tumor survivors and their families [21,33,47]. Survivors were identified through the Swedish Childhood Cancer Registry comprising information about primary cancer diagnosis, classified according to the International Classification for Childhood Cancer [48]. Parents were eligible for the study if their child was diagnosed with a primary CNS tumor between 1982 and 2001, <19 years of age at diagnosis, ≥ 18 years of age at time of follow-up, and for whom >5 years had elapsed from diagnosis to follow-up. Of 5443 children diagnosed with cancer in 1982–2001, 1535 (28%) had a primary CNS tumor. At the time of study, 460 of them died, and 697 survivors fulfilled inclusion criteria. Twenty-two parents of these could not be reached. Among the remaining participants, 556 completed the questionnaire. Responding ($n = 556$) and non-responding ($n = 119$) parents did not differ regarding diagnosis, child's sex, age at diagnosis, time since diagnosis, and age at follow-up (child and parent). After the exclusion of five incomplete questionnaires, data from 551 parents (81.6%) remained for analysis. Parents were instructed to complete the questionnaires together when possible. The characteristics of the study group are presented in Table 1. After approval from the Regional Research Ethics Committee and informed consent from all the participants, data were collected from June 2006 to March 2007. Figure 1 presents the study process.

Assessments

Impact on family scale

The Impact on Family Scale (IFS) addresses parent-perceived impact of their child's condition on families' everyday life, including subjective psychological burden and concrete issues about economy, planning needs, and opportunities to travel. It was chosen because it was seen to provide knowledge about how families of adult survivors need to make changes in their life situation and about targets of implementable interventions. The domains covered by the IFS are based on what was found relevant after conducting patient interviews, reviewing the literature, and applying clinically attained knowledge about patients with chronic conditions. This conceptual framework provides the justification of the addressed domains [46]. It has been used for the study of various diagnostic groups, including childhood cancer [9,10,37,38,49].

The 33 items address family impact in five domains: *social and familial disruption*, *personal strain*, *financial burden*, *mastery*, and *sibling impact*. Responses to statements (items) are given on a 4-point Likert scale, alternatives ranging from *strongly agree* to *strongly disagree*. The mastery domain was disregarded because of its unsatisfactory psychometric features [49], and a revised and improved scoring algorithm was used [49,50]. The IFS provides a separate outcome for each domain and a total impact score based on two domains

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Table 1. Study group characteristics

	Parents ($n = 551$)
Parent factors	
Responder—*n* (%)	
Mother	406 (73.7)
Father	31 (5.6)
Parents together	111 (20.1)
Missing	3 (0.5)
Age at assessment (years)	
Mean (SD)	54.0 (6.4)
n (%)	
<45	40 (7.3)
45–54	275 (49.9)
55–65	205 (37.2)
>65	30 (5.4)
Missing	1 (0.2)
Child factors	
Sex—*n* (%)	
Female	258 (46.8)
Male	293 (53.2)
Age at assessment (years)	
Mean (SD)	25.9 (5.0)
n (%)	
<25	270 (49.0)
25–30	183 (33.2)
31–36	78 (14.2)
>36	20 (3.6)
Age at diagnosis (years)	
Mean (SD)	10.3 (4.5)
Time since diagnosis (years)	
Mean (SD)	15.8 (5.1)
n (%)	
<10	100 (18.1)
10–15	140 (25.4)
>15–20	179 (32.5)
>20	132 (24.0)
Diagnosis—*n* (%)	
Astrocytoma	252 (45.7)
Ependymoma	52 (9.4)
Medulloblastoma/PNET	68 (12.3)
Other CNS tumors	179 (32.5)
History of relapse—*n* (%)	
Relapse	68 (12.3)
No relapse	483 (87.7)

SD, standard deviation; PNET, primitive neuroectodermal tumor; CNS, central nervous system.

(personal strain and familial/social disruption). Higher scores correspond to a greater adverse impact. Mean scores were used for outcomes, which can be presented as either sum or mean scores [8,10,36,37].

In this study, the internal reliability estimated by Cronbach's alpha ranged from 0.78 (sibling impact) to 0.92 (total impact), indicating high internal consistency.

Health-care needs

Parents reported on their child's health-care needs in adult life by completing an 11-item questionnaire used in prior studies of satisfaction with health-care services [8,51]. It covers four domains related to *medical care, care coordination/communication*, *illness education*, and *psychosocial/social counseling*. For each item, parents indicated whether the survivor had (i) received the particular service; (ii) received it partly; (iii) did not receive it and had no need for it; or (iv) did not receive

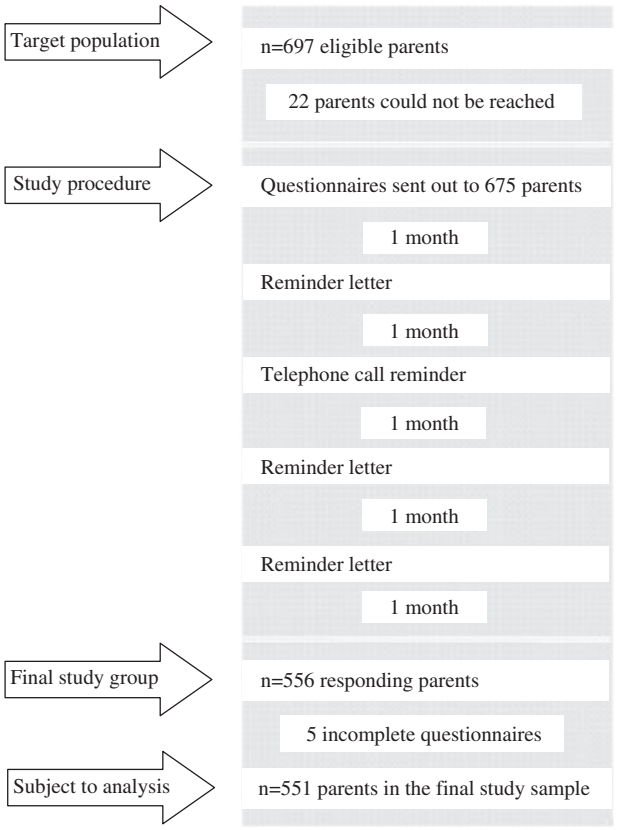

Figure 1. Study process

it although there was a need for it. An unmet health-care need was considered to be present for those who responded positively regarding the fourth alternative [8]. An unmet health-care need in a particular domain was considered present if at least one defined need (item) was unmet. For the psychosocial/social counseling domain, comprising a greater number of items than other domains, at least two defined needs had to be unmet for an unmet need to be recorded.

Satisfaction with information

Parents' satisfaction with illness/treatment-related information provided during treatment and follow-up was assessed using a summary item of a European Organisation for Research and Treatment of Cancer (EORTC) QOL-INFO26-based questionnaire [52]. For this summary item, parents indicated whether they were (i) *not satisfied*; (ii) *somewhat satisfied*; (iii) *satisfied to a great deal*; or (iv) *very satisfied* with the information they had been provided.

Health status

The Health Utilities Index^{TM}, Mark 2/3 (HUI) was used to study the extent to which survivors' current health status was associated with family impact [53]. The HUI is applicable to a variety of populations, including childhood CNS tumor survivors [33,54,55]. The overall health status outcome of the HUI was used, which defines health on an interval scale where 1.00 = *perfect*

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health and 0.00 = *dead*. Health status is categorized as indicating *mild disability* (0.89–0.99), *moderate disability* (0.70–0.88), or *severe disability* (<0.70) [53,54,56]. Parent-reported health data were used in this study. Detailed health outcomes for the study population have been presented previously [33].

Statistical analysis

Responders/non-responders differences were analyzed using *t*-tests for independent groups and chi-square tests.

Descriptive summary statistics are presented for family impact (IFS) outcomes. Scores equal to or greater than 2.5 were considered as indicating a significant impact. The cutoff for the significant impact was rationally based on how the response alternatives to the questionnaire items/questions were expressed; on the 4-point scale, alternatives 1 and 2 (representing a score value of 2 or below) denoted no impact, whereas alternatives 3 and 4 denoted experienced impact.

The associations between possible modifying background factors and IFS outcomes were analyzed using independent analysis of variance (ANOVA) and *t*-test, that is, in relation to parent responder constellation (whether the mother, father, or both provided data); age of parent and survivors at assessment; treatment center; and survivor's sex.

The influences of the determinants of primary interest were analyzed using ANOVA for independent and dependent samples and *t*-test, that is, IFS outcomes in relation to time since diagnosis; age at diagnosis; survivor's health status; relapse; parent-perceived unmet health-care needs; illness information satisfaction; and distance to specialized center. Dependent sample tests were conducted when analyses involved separate data sets that both were parent reported.

In final adjusted multivariable analyses of variance, the modifying factors that in preceding analyses were found to be significantly associated with family impact were inserted in the model to determine main and interaction effects. Interaction effects were evaluated only for factors for which a main effect for the IFS outcomes had been verified in the multivariable model.

Two-tailed testing was applied. To adjust for the use of multiple tests, we chose an alpha level of $p < 0.01$ rather than $p < 0.05$ as the threshold for statistical significance [57]. Analyses were carried out using SPSS version 17.0 (SPSS Inc., Chicago, IL).

Results

Because the outcomes did not significantly differ for parent responder constellations and treatment sites, the data from all respondents and sites were merged for analyses.

Impact on family

Although the impact on the families appeared to be mild to moderate at the group level (Table 2), only 6.5% of families reported no persistent impact whatsoever. In single domains, the corresponding proportions of parents reporting no impact varied between 17% and 32% (Table 2). The proportion of parents reporting a significant impact (scores ≥2.5) varied with domain. The greatest impact was found for *personal strain* and *financial burden*, where 18–20% of parents reported a significant impact, followed by *sibling-related impact* (11%) and *social/familial disruption* (7%) (Table 2).

Modifying factors

Family impact was associated with survivors' current health (Table 3). The illness-related impact on families of survivors with compromised health was significantly greater than in those of survivors with mild or no health-related sequelae. ANOVA followed by a paired *t*-test procedure with Bonferroni correction confirmed this pattern. Differences confirming the effect of impaired health were statistically significant in all comparisons, except when families of survivors of perfect health and survivors with mild disability were compared and when the moderate disability and severe disability groups were contrasted regarding sibling-related impact. Families of female survivors reported greater impact than families of male survivors (Table 3). Also, the impact was greater in families where survivors were perceived as having unmet health-care needs compared with when such needs were perceived as fully or partly met (Table 4). Forty-eight percent of parents reported no satisfaction ($n = 91$)

Table 2. Family impact scores and proportions (%) of families by amount of impact

		Amount of impact						
	Parents	No impact	Mild impact		Moderate impact		Great impact	
	n = 551	1.00	1.01–1.49	1.50–1.99	2.00–2.49	2.50–2.99	3.00–3.49	3.50–4.00
Impact domain	**Mean (SD)**	***n* (%)**	***n* (%)**	***n* (%)**	***n* (%)**	***n* (%)**	***n* (%)**	***n* (%)**
Personal strain	1.80 (0.69)	127 (23.0)	77 (14.0)	106 (19.2)	133 (24.1)	69 (12.5)	28 (5.1)	11 (2.0)
Social/familial disruption	1.45 (0.54)	169 (30.7)	207 (37.6)	66 (12.0)	71 (12.9)	31 (5.6)	6 (1.1)	1 (0.2)
Financial burden	1.77 (0.75)	178 (32.3)	67 (12.2)	71 (12.9)	136 (24.7)	52 (9.4)	36 (6.5)	11 (2.0)
Sibling impact[a]	1.70 (0.58)	83 (18.3)	91 (20.1)	108 (23.8)	122 (26.9)	37 (8.2)	9 (2.0)	3 (0.7)
Total impact	1.59 (0.56)	95 (17.2)	206 (37.4)	117 (21.2)	88 (16.0)	32 (5.8)	11 (2.0)	2 (0.4)

SD, standard deviation.

[a]Relevant only in families with a sibling ≥4 years old at follow-up ($n = 453$).

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Table 3. Family impact scores by survivor's sex and health status

	Survivor's sex		Survivor's health status			
	Male ($n = 293$)	Female ($n = 258$)	Perfect health ($n = 115$)	Mild disability ($n = 175$)	Moderate disability ($n = 114$)	Severe disability ($n = 126$)
Impact domain	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**
Personal strain	1.74 (0.65)	1.87 (0.74)	1.43 (0.53)	1.62 (0.63)	1.94 (0.62)	2.26 (0.66)**
Social/familial disruption	1.39 (0.48)	1.52 (0.59)**	1.19 (0.33)	1.28 (0.40)	1.50 (0.47)	1.88 (0.65)**
Financial burden	1.69 (0.69)	1.87 (0.79)**	1.44 (0.60)	1.60 (0.61)	1.83 (0.72)	2.23 (0.81)**
Sibling impact[a]	1.63 (0.55)	1.79 (0.61)**	1.43 (0.45)	1.58 (0.56)	1.81 (0.54)	2.00 (0.58)**
Total impact	1.53 (0.50)	1.66 (0.61)**	1.29 (0.37)	1.41 (0.44)	1.68 (0.48)	2.03 (0.61)**

[a]Relevant only in families with a sibling ≥4 years old ($n = 453$) at follow-up.
*$p < 0.01$, **$p < 0.001$ = significance level for differences due to sex, and due to health status of survivor.

Table 4. Family impact scores by parent-perceived unmet health-care needs of their child[a]

	Medical care		Care coordination		Illness education		Psychosocial services		Total	
	Unmet needs $n = 82$	No unmet needs $n = 306$	Unmet needs $n = 104$	No unmet needs $n = 297$	Unmet needs $n = 143$	No unmet needs $n = 238$	Unmet needs $n = 139$	No unmet needs $n = 160$	Unmet needs $n = 219$	No unmet needs $n = 251$
Impact domain	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**
Personal strain	2.07 (0.81)	1.91 (0.66)	2.10 (0.73)	1.85 (0.68)	2.18 (0.72	1.83(0.65)**	2.22 (0.71)	2.04 (0.60)	2.08 (0.70)	1.70 (0.64)**
Social/familial disruption	1.72 (0.69)	1.51 (0.52)	1.70 (0.61)	1.48 (0.54)	1.72 (0.62)	1.47(0.51)**	1.70 (0.60)	1.65 (0.56)	1.62 (0.59)	1.39 (0.50)**
Financial burden	2.05 (0.87)	1.89 (0.72)	2.04 (0.72)	1.83 (0.76)	2.10 (0.77)	1.81(0.73)**	2.08 (0.76)	2.02 (0.74)	2.00 (0.77)	1.71 (0.71)**
Sibling impact[b]	1.86 (0.60)	1.79 (0.59)*	1.91 (0.61)	1.75 (0.57)*	1.98 (0.62)	1.73(0.55)*	2.03 (0.62)	1.85 (0.54)	1.90 (0.61)	1.64 (0.53)**
Total impact	1.86 (0.70)	1.67 (0.53)	1.86 (0.61)	1.63 (0.55)*	1.90 (0.61)	1.61(0.52)**	1.91 (0.60)	1.81 (0.53)	1.81 (0.58)	1.51 (0.51)**

[a]Unmet/no unmet needs among those who reported having a health-care need to begin with.
[b]Relevant only in families with a sibling ≥4 years old ($n = 453$) at follow-up.
*$p < 0.01$, **$p < 0.001$ = significance level for differences between those with, and with no, unmet needs.

or only minor satisfaction ($n = 167$) with the illness-related information provided during treatment and follow-up. These 258 parents reported a greater family impact than the 279 parents reporting moderate ($n = 201$) or great ($n = 78$) satisfaction with information ($p < 0.001$ in all domains). However, the impact was unrelated to survivor's current age, age at diagnosis, parent's age, and distance to specialized treatment center (≤50 km or >50 km). Parents of survivors with a relapse history presented a higher, although statistically non-significant, impact in all domains. Time since diagnosis was related only to the social and familial disruption domains, with longer time being associated with greater impact ($F[3,547] = 4.704$, $p = 0.003$).

Multivariable analyses

In multivariable models, *survivor's sex* was not significantly associated with any of the IFS outcomes at the applied $p < 0.01$ level, whereas *survivors' health* remained significantly associated with all IFS outcomes (Table 5).

Parental satisfaction with information remained significantly influential in adjusted analyses regarding total impact, personal strain, and sibling-related impact. The significant ($p < 0.01$) influence of parental satisfaction with information established in unadjusted analyses disappeared regarding the domains of social and familial disruption ($p = 0.048$) and financial impact ($p = 0.115$).

In adjusted analyses, *parental perceived unmet (total) health-care needs* remained significantly associated with total impact, personal strain, and financial impact. The influence of *perceived unmet health-care needs* was reduced to below the $p < 0.01$ level regarding social and familial disruption ($p = 0.039$) and sibling-related impact ($p = 0.014$).

The significant influence established in unadjusted analyses regarding *time elapsed from diagnosis* and social and familial disruption disappeared in the adjusted analysis ($p = 0.265$).

No significant interaction effects were noticed.

Discussion

This study systematically evaluated the illness-related impact on families of adult survivors of childhood CNS tumors, a vulnerable group previously not investigated with this aim in view. The population-based data from an unusually large sample strengthens the reliability of the findings. At group level, the conditions of families of adult CNS tumor survivors appeared as mildly to moderately influenced by their child's past illness. However, in a considerable subgroup of parents, the illness-related impact persists and extends to several

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Table 5. Summary of main effects found in multivariable analyses of potentially family impact-influencing variables[a]

Impact domain	Survivors' sex	Survivors' health	Information satisfaction	Perceived unmet health-care needs	Time from diagnosis
Personal strain	—	$p < 0.01$	$p < 0.01$	$p < 0.01$	—
Social/familial disruption	ns	$p < 0.01$	ns	ns	ns
Financial burden	ns	$p < 0.01$	ns	$p < 0.01$	—
Sibling impact	ns	$p < 0.01$	$p < 0.01$	ns	—
Total impact	ns	$p < 0.01$	$p < 0.01$	$p < 0.01$	—

ns, not significant.
[a]Analyses conducted separately for each domain.
— Not included, as the variable was unrelated to the particular domain in initial univariable analyses.

domains of life. About one fifth of parents reported a persistent influence not only on personal well-being but also on family finances. Only a minor portion of families (6.5%) reported no remaining impact whatsoever at this late stage of follow-up.

In previous studies where the same family impact measure (IFS) has been used, families of younger childhood cancer patients both on and off treatment have reported a greater impact than found in our study [36–38], indicating, as expected, a greater impact of illness in caring for a younger child or one undergoing active treatment. Interestingly, we found no relationship between the strength of impact and the time passed since diagnosis or survivors' age, indicating that the adverse illness-related impact that some families of adult survivors experience tends to persist over time.

Consistent with prior studies of younger children suffering from disability [8,58], the greatest impact was established for the personal strain domain that relates to the heightened demands because of the consequences of the illness. On an item-level, the kind of strain reported as most strongly occurring was the heightened sensitivity parents felt to shifts in the well-being of the child (item-level analyses not presented).

Childhood cancer has previously, in studies of mixed diagnostic childhood cancer groups, been verified as a source of financial strain in families [59,60]. A remarkable finding in the present study was that families reported adverse financial consequences even after children have reached adulthood. The Swedish welfare system guarantees free medical care for children in a fairly equitable manner. Financial aid is available for families of children with chronic medical conditions demanding extraordinary care. Against that background, our finding that one fifth of parents continually experience financial difficulties was unexpected. The adverse financial consequences mainly resulted from medical care visits interfering with work and also from reduced paid job hours.

Prior studies have shown a substantial impact on social relationships/activities within the family and with others [8,37]. The finding that such consequences were rarely reported in our study indicates that families might have gradually catched up on such activities later on.

The findings regarding examined impact-modifying factors suggest that the persistent impact of illness was greatest in families of survivors with impaired health, thus supporting what has previously been indicated on the basis of smaller studies of families of younger clinical populations [9,10,61]. Our study shows that families of CNS tumor survivors, who are more likely to face sequelae than other pediatric patient groups, still suffer persistent adverse consequences even after the child reaches adulthood. The difficulties and the concerns of survivors have been found to be important determinants of parental worry in families of young survivors [61]. Such observations seem to correspond with our findings, as parents experiencing unmet health-care needs of their child—indicative of difficulties—reported a greater adverse impact on the family compared with families where such unmet needs were not a recognized problem. Unmet health-care needs have been considered as specifically contributing to the burden on families of children suffering from chronic conditions who are closer to diagnosis [8]. Our findings demonstrate that such perceived unmet needs add burden on families of adult childhood CNS tumor survivors as well. Unmet informational needs were also associated with the persistent adverse impact on the family. Whereas, again, similar findings have been presented for families of younger patients closer to the onset of child's cancer [19,62], our findings show that the importance of illness-related information is apparent also in adult survivor families.

The findings from this study have obvious implications for the surveillance of patients and long-term family care for this particularly vulnerable group. First, findings reveal the need for establishing specialized family support and counseling for unlimited time, that is, irrespective of age of the patient and time that has passed since illness and treatment. Second, patients' lingering late effects and unmet health-care requirements, which place a burden on the family, indicate the need for establishing effective clinical surveillance combined with support measures within the general welfare system to meet the specific needs of CNS tumor survivors requiring long-term health care.

This study has some limitations. First is the fact that the unmet health-care needs of survivors were parent reported and not supplemented with other sources of information. Second, because of the illness-specific nature of the family impact measure, we could not compare outcomes with what might be associated with parenthood in general. The IFS is an illness-specific measure, addressing unique problems of families who encountered a serious/chronic childhood illness—thus

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complicating the use of a comparison group. Third, the cross-sectional design prevents us from knowing whether observed family impact is a persistent or late-occurring phenomenon. The lack of additional socio-demographic characteristics of parents also constitutes a limitation. It prevents us to evaluate how such parent-related factors might be related to family impact. Finally, reliance on an impact questionnaire with predetermined domains may result in other potentially important family consequences, such as a specific impact on one's relationship/marriage and care responsibilities, remain unexplored.

Forthcoming research could address the long-term family impact using expanded data sources, including the use of additional impact measures to explore other relevant areas of a long-term impact. Mothers', fathers', and siblings' unique experiences constitute other targets of future studies for expanded knowledge about the consequences of childhood cancer in the family.

Conclusions

This study of parents of adult survivors of childhood CNS tumors presents evidence of persistent adverse family consequences in multiple domains. Depending on domain, we identified the significant adverse impact in 7–20% of parents. Impact was associated with the current health of survivors, unsatisfied informational needs, and parental experience of their adult child's unmet health-care needs. The adverse family impact could therefore be reduced by providing information tailored to the needs of families of CNS tumor patients, counseling on the likelihood of late effects in the individual survivor, and an added focus on health-care needs in adult life. These measures should complement the general welfare system when access to hospital-based surveillance and extended specialized follow-up is not available.

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References

1. Hoekstra-Weebers JE, Jaspers JP, Kamps WA, Klip EC. Psychological adaptation and social support of parents of pediatric cancer patients: a prospective longitudinal study. *J Pediatr Psychol* 2001;**26**:225–35.
2. Maurice-Stam H, Oort FJ, Last BF, Grootenhuis MA. Emotional functioning of parents of children with cancer: the first five years of continuous remission after the end of treatment. *Psycho-Oncology* 2008;**17**:448–59.
3. Boman KK, Viksten J, Kogner P, Samuelsson U. Serious illness in childhood: the different threats of cancer and diabetes from a parent perspective. *J Pediatr* 2004;**145**:373–9.
4. Kazak AE, Barakat LP, Meeske K *et al*. Posttraumatic stress, family functioning, and social support in survivors of childhood leukemia and their mothers and fathers. *J Consult Clin Psychol* 1997;**65**:120–9.
5. Kupst MJ, Natta MB, Richardson CC, Schulman JL, Lavigne JV, Das L. Family coping with pediatric leukemia: ten years after treatment. *J Pediatr Psychol* 1995;**20**:601–17.
6. Norberg AL, Lindblad F, Boman KK. Parental traumatic stress during and after paediatric cancer treatment. *Acta Oncol* 2005;**44**:382–8.
7. Peterson CC, Drotar D. Family impact of neurodevelopmental late effects in survivors of pediatric cancer: review of research, clinical evidence, and future directions. *Clin Child Psychol Psychiatry* 2006;**11**:349–66.
8. Thyen U, Sperner J, Morfeld M, Meyer C, Ravens-Sieberer U. Unmet health care needs and impact on families with children with disabilities in Germany. *Ambul Pediatr* 2003;**3**:74–81.
9. Ireys HT, Silver EJ. Perception of the impact of a child's chronic illness: does it predict maternal mental health? *J Dev Behav Pediatr* 1996;**17**:77–83.
10. Schwartz L, Taylor HG, Drotar D, Yeates KO, Wade SL, Stancin T. Long-term behavior problems following pediatric traumatic brain injury: prevalence, predictors, and correlates. *J Pediatr Psychol* 2003;**28**:251–63.
11. Hovén E, Anclair M, Samuelsson U, Kogner P, Boman KK. The influence of pediatric cancer diagnosis and illness complication factors on parental distress. *J Pediatr Hematol Oncol* 2008;**30**:807–14.
12. Yantzi N, Rosenberg MW, Burke SO, Harrison MB. The impacts of distance to hospital on families with a child with a chronic condition. *Soc Sci Med* 2001;**52**:1777–91.
13. Litzelman K, Catrine K, Gangnon R, Witt WP. Quality of life among parents of children with cancer or brain tumors: the impact of child characteristics and parental psychosocial factors. *Qual Life Res* 2011, DOI: 10.1007/s11136-011-9854-2
14. Witt WP, Litzelman K, Wisk LE *et al*. Stress-mediated quality of life outcomes in parents of childhood cancer and brain tumor survivors: a case-control study. *Qual Life Res* 2010;**19**:995–1005.
15. Reiter-Purtill J, Schorry EK, Lovell AM, Vannatta K, Gerhardt CA, Noll RB. Parental distress, family functioning, and social support in families with and without a child with neurofibromatosis 1. *J Pediatr Psychol* 2008;**33**:422–34.
16. Norberg AL, Steneby S. Experiences of parents of children surviving brain tumour: a happy ending and a rough beginning. *Eur J Cancer Care (Engl)* 2009;**18**:371–80.
17. Eiser C. Practitioner review: long-term consequences of childhood cancer. *J Child Psychol Psychiatry* 1998;**39**: 621–33.
18. Kinahan KE, Sharp LK, Arntson P, Galvin K, Grill L, Didwania A. Adult survivors of childhood cancer and their parents: experiences with survivorship and long-term follow-up. *J Pediatr Hematol Oncol* 2008;**30**:651–8.
19. Freeman K, O'Dell C, Meola C. Childhood brain tumors: parental concerns and stressors by phase of illness. *J Pediatr Oncol Nurs* 2004;**21**:87–97.
20. Vance YH, Eiser C, Horne B. Parents' views of the impact of childhood brain tumours and treatment on young people's social and family functioning. *Clin Child Psychol Psychiatry* 2004;**9**:271–288.
21. Anclair M, Hovén E, Lannering B, Boman KK. Parental fears following their child's brain tumor diagnosis and treatment. *J Pediatr Oncol Nurs* 2009;**26**:68–74.
22. Young B, Dixon-Woods M, Findlay M, Heney D. Parenting in a crisis: conceptualising mothers of children with cancer. *Soc Sci Med* 2002;**55**:1835–47.
23. Gannoni AF, Shute RH. Parental and child perspectives on adaptation to childhood chronic illness: a qualitative study. *Clin Child Psychol Psychiatry* 2010;**15**:39–53.

24. Coffey JS. Parenting a child with chronic illness: a metasynthesis. *Pediatr Nurs* 2006;**32**:51–9.
25. Kazak AE. Families of chronically ill children: a systems and social-ecological model of adaptation and challenge. *J Consult Clin Psychol* 1989;**57**:25–30.
26. Ressler IB, Cash J, McNeill D, Joy S, Rosoff PM. Continued parental attendance at a clinic for adult survivors of childhood cancer. *J Pediatr Hematol Oncol* 2003;**25**: 868–73.
27. Mueller S, Chang S. Pediatric brain tumors: current treatment strategies and future therapeutic approaches. *Neurotherapeutics* 2009;**6**:570–86.
28. Gustafsson G, Heyman M, Vernby Å, Childhood cancer incidence and survival in Sweden 1984–2005. In *Report from the Swedish Childhood Cancer Registry*. 2007, Karolinska Institutet: Stockholm.
29. Mulhern RK, Merchant TE, Gajjar A, Reddick WE, Kun LE. Late neurocognitive sequelae in survivors of brain tumours in childhood. *Lancet Oncol* 2004;**5**:399–408.
30. Lannering B, Marky I, Nordborg C. Brain tumors in childhood and adolescence in west Sweden 1970–1984. Epidemiology and survival. *Cancer* 1990;**66**:604–9.
31. Oeffinger KC, Mertens AC, Sklar CA *et al*. Chronic health conditions in adult survivors of childhood cancer. *N Engl J Med* 2006;**355**:1572–82.
32. Hjern A, Lindblad F, Boman KK. Disability in adult survivors of childhood cancer: a Swedish national cohort study. *J Clin Oncol* 2007;**25**:5262–6.
33. Boman KK, Hovén E, Anclair M, Lannering B, Gustafsson G. Health and persistent functional late effects in adult survivors of childhood CNS tumours: a population-based cohort study. *Eur J Cancer* 2009;**45**:2552–61.
34. Blaauwbroek R, Stant AD, Groenier KH, Kamps WA, Meyboom B, Postma A. Health-related quality of life and adverse late effects in adult (very) long-term childhood cancer survivors. *Eur J Cancer* 2007;**43**:122–30.
35. Fuemmeler BF, Mullins LL, Marx BP. Posttraumatic stress and general distress among parents of children surviving a brain tumor. *Child Health Care* 2001;**30**:169–182.
36. Bonner MJ, Hardy KK, Willard VW, Hutchinson KC. Brief report: psychosocial functioning of fathers as primary caregivers of pediatric oncology patients. *J Pediatr Psychol* 2007;**32**:851–6.
37. Heath JA, Lintuuran RM, Rigguto G, Tikotlian N, McCarthy M. Childhood cancer: its impact and financial costs for Australian families. *Pediatr Hematol Oncol* 2006;**23**:439–48.
38. Sawyer M, Antoniou G, Toogood I, Rice M. A comparison of parent and adolescent reports describing the health-related quality of life of adolescents treated for cancer. *Int J Cancer Suppl* 1999;**12**:39–45.
39. Foley B, Barakat LP, Herman-Liu A, Radcliffe J, Molloy P. The impact of childhood hypothalamic/chiasmatic brain tumors on child adjustment and family functioning. *Child Health Care* 2000;**29**:209–223.
40. Patterson JM, Holm KE, Gurney JG. The impact of childhood cancer on the family: a qualitative analysis of strains, resources, and coping behaviors. *Psycho-Oncology* 2004;**13**:390–407.
41. Stehl ML, Kazak AE, Alderfer MA *et al*. Conducting a randomized clinical trial of an psychological intervention for parents/caregivers of children with cancer shortly after diagnosis. *J Pediatr Psychol* 2009;**34**:803–16.
42. Hardy KK, Bonner MJ, Masi R, Hutchinson KC, Willard VW, Rosoff PM. Psychosocial functioning in parents of adult survivors of childhood cancer. *J Pediatr Hematol Oncol* 2008;**30**:153–9.
43. Doshi K, Kazak AE, Derosa BW *et al*. Measuring health-related beliefs of mothers of adolescent and young adult childhood cancer survivors. *Fam Syst Health* 2011;**29**:55–63.
44. Jackson AC, Tsantefski M, Goodman H, Johnson B, Rosenfeld J. The psychosocial impacts on families of low-incidence, complex conditions in children: the case of craniopharyngioma. *Soc Work Health Care* 2003;**38**:81–107.
45. Jackson AC, Enderby K, O'Toole M *et al*. The role of social support in families coping with childhood brain tumor. *J Psychosoc Oncol* 2009;**27**:1–24.
46. Stein RE, Riessman CK. The development of an impact-on-family scale: preliminary findings. *Med Care* 1980; **18**:465–72.
47. Hovén E, Lannering B, Gustafsson G, Boman KK. The met and unmet health care needs of adult survivors of childhood central nervous system tumors: a double-informant, population-based study. *Cancer* 2011;**117**:4294–4303.
48. Steliarova-Foucher E, Stiller C, Lacour B, Kaatsch P. International classification of childhood cancer, third edition. *Cancer* 2005;**103**:1457–67.
49. Stein RE, Jessop DJ. The impact on family scale revisited: further psychometric data. *J Dev Behav Pediatr* 2003; **24**:9–16.
50. Williams AR, Piamjariyakul U, Williams PD, Bruggeman SK, Cabanela RL. Validity of the revised Impact on Family (IOF) scale. *J Pediatr* 2006;**149**:257–61.
51. Stein RE, Jessop DJ. Does pediatric home care make a difference for children with chronic illness? Findings from the Pediatric Ambulatory Care Treatment Study. *Pediatrics* 1984;**73**:845–53.
52. Arraras JI, Kuljanic-Vlasic K, Bjordal K *et al*. EORTC QLQ-INFO26: a questionnaire to assess information given to cancer patients a preliminary analysis in eight countries. *Psycho-Oncology* 2007;**16**:249–54.
53. Horsman J, Furlong W, Feeny D, Torrance G. The Health Utilities Index (HUI(R)): concepts, measurement properties and applications. *Health Qual Life Outcomes* 2003;**1**:54.
54. Furlong WJ, Feeny DH, Torrance GW, Barr RD. The Health Utilities Index (HUI) system for assessing health-related quality of life in clinical studies. *Ann Med* 2001;**33**:375–84.
55. Glaser A, Kennedy C, Punt J, Walker D. Standardized quantitative assessment of brain tumor survivors treated within clinical trials in childhood. *Int J Cancer Suppl* 1999;**12**:77–82.
56. Feeny D, Farris K, Cote I, Johnson JA, Tsuyuki RT, Eng K. A cohort study found the RAND-12 and Health Utilities Index Mark 3 demonstrated construct validity in high-risk primary care patients. *J Clin Epidemiol* 2005;**58**:138–41.
57. Veazie PJ. When to combine hypotheses and adjust for multipletests. *Health Serv Res* 2006;**41**:804–18.
58. Kramer FJ, Baethge C, Sinikovic B, Schliephake H. An analysis of quality of life in 130 families having small children with cleft lip/palate using the impact on family scale. *Int J Oral Maxillofac Surg* 2007;**36**:1146–52.
59. Long KA, Marsland AL. Family adjustment to childhood cancer: a systematic review. *Clin Child Fam Psychol Rev* 2011;**14**:57–88.
60. Tsimicalis A, Stevens B, Ungar WJ, McKeever P, Greenberg M. The cost of childhood cancer from the family's perspective: a critical review. *Pediatr Blood Cancer* 2011; **56**:707–17.
61. Zebrack BJ, Chesler M, Orbuch TL, Parry C. Mothers of survivors of childhood cancer: their worries and concerns. *J Psychosoc Oncol* 2002;**20**:1–25.
62. Trask CL, Welch JJ, Manley P, Jelalian E, Schwartz CL. Parental needs for information related to neurocognitive late effects from pediatric cancer and its treatment. *Pediatr Blood Cancer* 2009;**52**:273–9.

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