The siblings of childhood cancer patients need early support: a follow up study over the first year

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Background and methods: In a 1 year follow up study, we assessed the life situation of 33 siblings of childhood cancer patients and 357 healthy controls. The hypothesis was that siblings have more behavioural and health related problems just after the cancer diagnosis. Validated assessment methods were used.

Results: Siblings below school age tended to have conduct problems, psychosomatic problems, and a mixed group of other behavioural problems, when assessed 3 months after the cancer diagnosis. These symptoms become less evident during follow up. Among the school aged siblings, however, conduct problems, learning problems, psychosomatic problems, impulsive-hyperactive symptoms, and other behavioural symptoms remained unchanged during follow up. In their self assessments, the school aged siblings showed both state and trait anxiety more often than controls at the first assessment, but later these symptoms settled to the same level as the controls. The overall Children’s Depression Inventory (CDI) depression scores did not show differences between the study groups.

Conclusions: The ratings of the parents were in keeping with the self assessment of the school aged siblings only in a few aspects; the emphasis of findings can be changed when proxies are used. The siblings have symptoms and adverse feelings which probably could be relieved by targeted, early information about the illness, and possibly by group discussions or activities, soon after the cancer diagnosis. In order to obtain necessary support for the siblings with educational problems, school personnel need to be informed about the sibling distress.

METHODS

The study took place in one of five Finnish centres treating childhood cancer, the University Hospital of Turku. The families of all patients diagnosed with cancer over a 15 month period were included. There were a total of 27 new patients, and 26 of their families participated (one family was not Finnish and was not eligible because of language problems). The characteristics of the study groups are presented in table 1. There were 70 control families for siblings below school age (<7 years) recruited from two regional day care centres (response rate 47%). Another group of healthy school aged (≥7 years) controls was recruited in two primary and two secondary schools. Informed consent was sought from the parents of students in one class in each of nine grades (n = 450), 74% of whom (n = 333) were allowed by their parents to participate. However, only 287 parents of these healthy controls returned their evaluation forms, so only children from these families were used in analyses. Two separate control groups were established to ensure the similarity of the family situations of families with cancer and controls, that is families with most children below school age and families with older children.

Abbreviations: CDI, Children’s Depression Inventory; CPRS-48, Conners’ Parent Rating Scales; SV, sum variables
Parents were asked to choose from among siblings below school age (aged 3–7 years) the child nearest the age of the patient, and fill in the questionnaire for that particular sibling. Additionally, all school aged siblings up to 17 years of age were assessed. We decided to assess all school aged siblings, instead of only one, because this would allow the attitudes of respondents and proxies to be compared.

Among the 26 study families, 13 had a diagnosis of leukaemia or lymphoma, seven a solid tumour, and six a brain tumour. The median (range) age of the patients at diagnosis was 4.5 (0.1–15) years. Five families were omitted from the follow up: four because of the unsuitable age (below 3 or over 17 years, which were the age limits for the standardised questionnaires used) of the siblings, and one because the family was not willing to participate further.

The median age (range) of the 12 below school age study siblings was 5.4 (3–7) years, while that of their 70 controls was 5 (3–7) years. The 21 school aged siblings were aged 10 (7–15) and their 287 controls were aged 11 (7–17). Some 16 of 21 school aged siblings were in primary school (grades 1–6).

**Measurements**

The first assessment of families with childhood cancer patients took place 3 months after cancer diagnosis and the second 12 months later. A questionnaire on background information and family life was given to the parents by one of the researchers, together with another questionnaire concerning siblings. For the school aged siblings, there were self assessment questionnaires and separate questionnaires filled in by their parents.

The questionnaires for the parents were: a validated Finnish test (by Huttunen) for behavioural assessment of kindergarten aged (>3–7 years) children\(^{17}\) (Appendix A) as well as Conners’ Parent Rating Scales (CPRS-48) for 3–17 years old children.\(^{18}\) These validated methods were chosen as their contents agree with findings reported in previous studies (see description below). The questionnaires were anonymous and were returned in closed envelopes to the investigators.

Huttunen’s test,\(^{17}\) which was originally modified from several recent studies in essential areas of child behaviour, has been successfully used, for example, in a nationwide analysis of the behaviour of Finnish children. The reliability of the questionnaire has been estimated by Cronbach’s coefficient \(\alpha\), regarding the evaluations of both parents and day nursery employees. The 46 multiple choice questions in this test assess aspects of the child’s behaviour to which parents pay attention, and deal with the child’s behaviour when he/she is alone, communicates with other children or adults, plays, or is involved in daily activities. Negative behavioural features and somatic symptoms are analysed in detail for possible conduct problems. The answers to each item are scored from 1 to 5 (very often, often, sometimes, seldom, never). In the sum variables (SV), negative behavioural features were marked with lower scores and positive behaviour with higher scores. The contents of the sum variables (see Appendix A) were as follows: SV1, behaviour in basic care situations; SV2, behaviour with other children; SV3, behaviour towards adults; SV4, behaviour in play activities; SV5, occurrence of somatic and psychosomatic symptoms; and SV6, confrontational situations with parents.---
Each of the 48 symptoms in Conners’ Parent Rating Scale was marked as not at all present, just a little present, pretty much present, or very much present, and scored from 0 to 3, respectively (a higher score indicating greater symptomatology). The items were grouped into conduct problems, learning problems, psychosomatic symptoms, impulsive-hyperactive behaviour, anxiety, and a mixed group of other behavioural problems.

In addition, three standardised self assessment forms were completed by the school aged siblings and their control group. The forms were: STAIC Form C-1 (20 questions) and STAIC Form C-2 (20 questions) to evaluate the anxiety and self esteem of the respondent (state and trait), and Childhood Depression Inventory (CDI) (32 questions) which evaluates the depression of the respondent during the past 2 weeks (see below). The questionnaires were given to the control students (as well as the CPRS-48 forms to be completed by their parents) at the beginning of a lesson, and the parental forms were anonymously mailed from home to the investigators.

STAIC Form C1, the A-State scale, is designed to measure transitory anxiety states, that is, subjective, consciously perceived feelings of apprehension, tension, and worry that vary in intensity and fluctuate over time. Form C2, the A-Trait scale, measures relatively stable individual differences in proneness to anxiety, that is, differences between children in their tendency to experience anxiety states. The CDI form mainly assesses the depressive aspects of a child’s behaviour. Total scores were calculated from each of the forms. However, we analysed the self assessments of school aged siblings also at the item level (in addition to the usual overall score comparisons) in order to detect which items mainly differentiate the study groups from each other.

Statistical analysis

Statistical analyses were performed using SAS for Windows, release 8.12. For descriptive statistics, medians and ranges were calculated, as the data were not normally distributed. Wilcoxon rank sum test (Mann-Whitney U) was used in analysing the numerical outcome variables, whereas the categorical variables were analysed with Fisher’s exact test. Because of the paired nature of the two assessments, Wilcoxon signed rank test was used when comparing the results of the first and second assessments of the siblings. Two sided p values of $<$0.05 were interpreted as statistically significant.

The Joint Commission on Ethics of Turku University Central Hospital accepted the research plan. The subjects and their guardians signed an informed consent form.

RESULTS

There were no statistically significant differences between the study group and the control group regarding age of the respondent, age of the study children, family size, or marital status of the parents. However, significantly more of the control respondents had a university degree. Also the monthly income was higher ($p = 0.03$ in the below school age group, $p = 0.001$ in the school aged group) among control families. However, this may have been due to the fact that the working status of the mothers of cancer patients differed significantly ($p = 0.022$) from the controls. At 3 months after the diagnosis of cancer, $33\%$ of the mothers stated that they had stopped working just because of the illness of their child, while a further $38\%$ were already at home because of maternity/child care leave, unemployment, or housewife status.

The results of the behavioural measures for siblings below school age are presented in table 2. The sum variables in Huttunen’s test showing statistically significant differences were “behaviour in play and activities” and “confrontational situations with parents”. At the first assessment, the siblings of cancer patients scored more negatively than the controls. However, the only statistically significant change ($p = 0.043$) in the behaviour of study siblings was in SV1 (behaviour in basic care situations). CPRS-48 showed a statistically significant difference between study and control siblings at the first assessment in the sum variable “other behavioural problems”, and in a trend in three sum variables, “conduct problems”, psychosomatic problems”, and “impulsive-hyperactive”. Impulsive-hyperactive behaviour tended to diminish in study siblings during follow up ($p = 0.09$).

The self assessment scores of the school aged siblings are listed in table 3. At the first assessment, state anxiety was greater in siblings than in controls ($p = 0.019$). This anxiety, however, diminished significantly during follow up ($p = 0.007$). In deeper analyses, it was revealed that this finding was more pronounced among the primary school siblings. The same trend was noticed in trait anxiety scores at the first assessment ($p = 0.056$). Neither significant differences nor trends were found in depression scores of the study subjects.

In the STAIC forms, the primary school aged siblings scored higher than controls in a few items, especially during the first assessment. These items were as follows: I feel very scared ($p = 0.0001$), I feel very frightened ($p = 0.041$), I feel not at all happy ($p = 0.03$), I feel very sad ($p = 0.009$), I feel very confused ($p = 0.0003$), I would like to cry often ($p = 0.044$), I feel it is difficult to face my problems ($p < 0.001$), Unnecessary thoughts often bother me ($p = 0.033$), I often feel my heart beating too fast ($p = 0.019$). The older siblings scored higher in the following items: I have difficulties falling asleep ($p = 0.017$), My hands sweat easily ($p = 0.062$).

In the CDI form, the siblings also scored higher than the controls in a few items, although the overall scores did not differ significantly. At the first assessment, the issues were as follows: Things never go well with me ($p = 0.049$ for the primary school siblings), I’m continuously worried about pains and aches ($p = 0.019$ for the primary school siblings), Nobody really likes me ($p = 0.038$ for the secondary school siblings). All the awful things are caused by me ($p = 0.044$ for the secondary school siblings). At the second assessment, the issues were as follows: I always have to force myself to do my homework ($p = 0.011$ for the primary school siblings), I can never be as good as the other children ($p = 0.032$ for the primary school siblings), I have sleeping difficulties each night ($p = 0.029$ for the secondary school siblings), I often have a headache ($p = 0.067$ for the secondary school siblings).

Appraisals of the parents of school aged siblings are presented in table 4. At the first assessment, conduct problems ($p = 0.002$) and other behavioural problems ($p < 0.001$) were reported significantly more in siblings than in controls. At the second assessment, all categories, except anxiety, showed significant score differences between controls and siblings. The changes in scores did not, however, reach statistical significance during follow up. When the parental assessments (CPRS-48) were evaluated in the primary school and secondary school age groups separately, the scores of secondary school siblings explained the differences in the overall scores: conductive problems ($p = 0.009$), learning problems ($p = 0.017$), psychosomatic problems ($p = 0.036$), and other behavioural problems ($p < 0.001$).

DISCUSSION

In this 1 year follow up study, we assessed the life situation of 33 siblings (12 below school age and 21 of school age) of
childhood cancer patients and 357 healthy controls. Despite a relatively small sample size, this study population represents an entire group of families of childhood cancer patients diagnosed over 15 months in one university hospital area.

The background variables of the study families and the controls were similar, except in terms of educational and working status. This finding may be due to the lower response rate of the control families using day care centres. There were more university graduates among the controls, but the response rate of the control families using day care centres. This finding may be due to the lower working status. This finding may be due to the lower response rate of the control families using day care centres. There were more university graduates among the controls, and a most mothers of cancer children stayed at home. These factors had a significant influence on the monthly income of the family. However, as social differences in Finland are narrow, health related issues would not be greatly affected by lower family income.

The Conners’ symptom rating scales (CPRS-48) were originally developed to help identify behavioural problems, such as anxiety, conduct problems, or hyperkinetic symptoms.8 We found that the siblings below school age when assessed soon after the cancer diagnosis tended to present with conduct problems, psychosomatic problems, impulsive-hyperactive behaviour, and a mixed group of other behavioural problems. All these symptoms became less evident during follow up, although statistical significance was not reached. The most notable change was the decrease in impulsive-hyperactive behaviour between the two assessments.

According to Huttunen’s scale,17 the siblings below school age had more problems soon after diagnosis than the controls, although the differences were statistically significant only as regards the items “behaviour in play and activities” and “confrontational situations with parents”. After 1 year follow up, the siblings scored similar to the controls, although paired analysis did not show any significant change. This finding is in keeping with the CPRS-48 results as the mentioned entities resemble each other.

The results of CPRS-48 agree with previous2–8 13 studies showing more behavioural and psychosomatic symptoms shortly after cancer diagnosis. Huttunen’s scale suggests minor problems. However, both tests showed that during follow up the symptom scores of the siblings improved. There are no previous data of similar follow up studies. Our findings agree with an earlier report showing that after cancer diagnosis the prevalence of parent reported emotional/behavioural problems among siblings is higher than in the general population.4 The results also support our hypothesis as the median scores improved.

The small number of problems reported by parents may be caused by lack of parental attention during the first months of treatment as suggested earlier.4 6 9 However, both tests suggested fewer problems existed after follow up. Thus, the parents did have at least some energy to pay also attention to

### Table 2 Results of the behavioural assessments in siblings and controls below school age

<table>
<thead>
<tr>
<th></th>
<th>Siblings (n = 12), median score (range)</th>
<th>Controls (n = 70), median score (range)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>At first assessment (I)</td>
<td>At second assessment (II)</td>
<td></td>
</tr>
<tr>
<td>Huttunen</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SV1 - Behaviour in basic care situations</td>
<td>14 (12–18)</td>
<td>16 (12–18)</td>
<td>1.5 (7–19)</td>
</tr>
<tr>
<td>SV2 - Behaviour with other children</td>
<td>22 (20–27)</td>
<td>23.5 (19–29)</td>
<td>25 (19–29)</td>
</tr>
<tr>
<td>SV3 - Behaviour towards adults</td>
<td>11.5 (9–16)</td>
<td>13 (9–17)</td>
<td>13 (9–19)</td>
</tr>
<tr>
<td>SV4 - Behaviour in play and activities</td>
<td>16 (12–21)</td>
<td>19.5 (10–25)</td>
<td>20 (12–28)</td>
</tr>
<tr>
<td>SV5 - Occurrence of somatic and psychosomatic symptoms</td>
<td>32 (15–29)</td>
<td>34 (23–37)</td>
<td>34 (19–43)</td>
</tr>
<tr>
<td>SV6 - Confrontational situations with parents</td>
<td>31 (23–45)</td>
<td>35 (27–43)</td>
<td>36 (20–44)</td>
</tr>
<tr>
<td>Conners’</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Conduct problems</td>
<td>5.5 (1–13)</td>
<td>3 (1–17)</td>
<td>2 (0–16)</td>
</tr>
<tr>
<td>Learning problems</td>
<td>1 (0–5)</td>
<td>1.5 (0–4)</td>
<td>1 (0–4)</td>
</tr>
<tr>
<td>Psychosomatic problems</td>
<td>1.5 (0–9)</td>
<td>1 (0–5)</td>
<td>0 (0–7)</td>
</tr>
<tr>
<td>Impulsive-hyperactive</td>
<td>4 (1–7)</td>
<td>1 (0–5)</td>
<td>3 (0–8)</td>
</tr>
<tr>
<td>Anxiety</td>
<td>0.5 (0–3)</td>
<td>1 (0–4)</td>
<td>1 (0–5)</td>
</tr>
<tr>
<td>Other behavioural problems</td>
<td>9 (4–27)</td>
<td>7 (2–12)</td>
<td>5 (0–24)</td>
</tr>
</tbody>
</table>

In the Huttunen scale, lower scores indicate more negative behaviour. In Conners’ scale, higher scores indicate more negative behaviour.

### Table 3 Total score results of the self assessments of school aged siblings and controls

<table>
<thead>
<tr>
<th></th>
<th>Siblings (n = 21), median score (range)</th>
<th>Siblings (n = 20), median score (range)</th>
<th>Controls (n = 238), median score (range)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>At first assessment (I)</td>
<td>At second assessment (II)</td>
<td></td>
<td>I v controls</td>
</tr>
<tr>
<td>STAIC Form C-1</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall</td>
<td>31.5 (27–40)</td>
<td>29.5 (20–40)</td>
<td>29 (20–52)</td>
<td>0.019</td>
</tr>
<tr>
<td>Primary school</td>
<td>31.5 (27–40)</td>
<td>29.5 (20–40)</td>
<td>27 (20–52)</td>
<td>0.003</td>
</tr>
<tr>
<td>Secondary school</td>
<td>31 (28–38)</td>
<td>30 (23–40)</td>
<td>30 (20–45)</td>
<td>0.54</td>
</tr>
<tr>
<td>STAIC Form C-2</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall</td>
<td>33.5 (23–44)</td>
<td>32 (20–48)</td>
<td>31.5 (20–53)</td>
<td>0.056</td>
</tr>
<tr>
<td>Primary school</td>
<td>33.5 (26–40)</td>
<td>32 (24–48)</td>
<td>30 (20–52)</td>
<td>0.054</td>
</tr>
<tr>
<td>Secondary school</td>
<td>33.5 (23–42)</td>
<td>31 (20–44)</td>
<td>31 (20–53)</td>
<td>0.30</td>
</tr>
<tr>
<td>CDI</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall</td>
<td>40 (34–56)</td>
<td>40.5 (33–60)</td>
<td>38 (32–67)</td>
<td>0.22</td>
</tr>
<tr>
<td>Primary school</td>
<td>39 (34–50)</td>
<td>40.5 (33–60)</td>
<td>38 (32–67)</td>
<td>0.25</td>
</tr>
<tr>
<td>Secondary school</td>
<td>41 (34–56)</td>
<td>40.5 (33–57)</td>
<td>37 (32–67)</td>
<td>0.62</td>
</tr>
</tbody>
</table>

In order to examine whether the overall effects of the trends noticed in total scores were mainly from the primary (n = 16) or the secondary (n = 5) school siblings, the data were analysed also over these subgroups of subjects. Higher scores indicate more distress.
the siblings. The family rehabilitation course might have had a positive effect on parental adaptation and family cohesion, which factors have earlier been reported to be associated with better adaptation of the siblings. On the other hand, the siblings may also get used to the changed family situation, their knowledge increasing as proposed earlier, or they may even want to protect their parents by behaving themselves.

The results of the CPRS-48 assessment of school aged siblings differed from those of the younger siblings. At follow up, there were significantly higher median scores for conduct problems, learning problems, psychosomatic problems, impulsive-hyperactive symptoms, and other behavioural symptoms. The self assessment results differed from parental opinion. The parents noticed more problems during follow up, whereas the siblings’ overall anxiety scores differed from controls only at the first assessment. This discrepancy in reports may be caused by lack of parental attention during the first months, as several researchers have suggested. On the other hand, the parents of cancer patients may, in the long run, be more sensitive to normal pubertal behaviour changes and get more irritated than the parents of healthy children. This result has not been reported previously. However, our findings stress the importance of several evaluation methods when assessing the distress of children by proxy respondents.

In their self assessments, the school aged siblings had higher overall scores on the STAIC at the first assessment. At the second assessment, these symptoms had more or less settled to the same level as the other study subjects. Our results suggest that previous studies stressing only the suffering of siblings below school age may be slightly misleading as school aged siblings also need support. A comprehensive list of guidelines for organising support for siblings is provided by the SIOP Working Committee, and the positive effect of a supportive group has been shown by Houtzager and co-workers.

The overall CDI scores indicating depression did not significantly differ between the study groups. However, there were tendencies to higher distress among the siblings when each of the questions was analysed separately. Our findings parallel previous reports in terms of school related problems and somatic complaints.

In conclusion, the CPRS-48 and Huttunen’s scale agree with previous studies showing more behavioural and psychosomatic symptoms in siblings below school age shortly after cancer diagnosis. The previous studies stressing only the suffering of siblings below school age may be slightly misleading as school aged siblings also showed symptoms. These findings parallel previous reports in terms of school related problems and somatic complaints, but no signs of social isolation or poor self esteem were seen. Our results highlight the problems with proxy respondents. As regards the siblings, parental appraisal is affected by their inability to concentrate on any other than the ill child, at least during the first months after the diagnosis of cancer. Thus, several methods of assessment are needed. Parents should also have an early opportunity to discuss with team members how to deal with questions and the possible problems of their other children. Siblings clearly have symptoms and adverse feelings which could perhaps be relieved by targeted, early information, and possibly by group discussions or activities, soon after the cancer diagnosis. Also, school personnel need to be informed about sibling distress.

APPENDIX A

ITEMS IN HUTTUNEN’S FORM (CHILDREN BELOW SCHOOL AGE)

A. Behaviour in basic care situations (sum variable 1)

- Independent in toilet.
- Dresses quickly.
- Restless and disturbed when eating.
- Dawdles over eating.
- Finds it difficult to settle down to sleep.
- Enjoys going to bed and falls asleep easily.

B. Behaviour with other children (sum variable 2)

- Takes into account the suggestions and activity of other children.
- Gives credit to playmates.
- Gives orders to other children.
- Argues with or teases other children, or disturbs their games.
- Fights with or hits other children.
- Is introvert and avoids the company of other children.
- Is timid and slow to make contacts with other children.
- Makes friends with children easily.

C. Behaviour toward adults (sum variable 3)

- Is enthusiastic about games and activity suggested by adults.
The siblings of cancer patients

- Denials and failures cause aggressive behaviour.
- Clings to adults and tries to monopolise their attention.
- Is timid and slow to make contact with adults.
- Makes friends with adults easily.

**D. Behaviour in play and activities (sum variable 4)**

- The child is active, creative, and shows initiative.
- Has a short attention span, finds it difficult to concentrate on games.
- Does not listen to what is said or advised.
- Gets carried away when playing.
- Becomes angry or loses his/her temper when fails or does not win.
- Is lively, always active, and enjoys playing with others.
- Is frustrated by failure and stops playing.

**E. Occurrences of (sum variable 5)**

- Bed wetting.
- Daytime wetting.
- Failures of bowel control.
- Headaches.
- Stomach aches.
- Running nose or cough.
- Fears.
- Anxiety.
- Sleeping problems.
- Nightmares.

**F. All parent face problems when raising their children. In which of the following situations have you found problems? (sum variable 6)**

- Going to bed or getting up.
- Eating.
- Disobedience or defiance.
- Looking after one’s things.
- Fighting with other children.
- Demands too much attention.
- Plays too roughly or noisily.
- Is irritable or bad tempered.
- Tantrums.
- Jealousies.

**REFERENCES**