CHRONIC FATIGUE IN SIBLINGS OF PEDIATRIC CANCER SURVIVORS: CHILDREN'S SELF-REPORTS AND PARENTS' PROXY-REPORTS

Likhomanova EN[1,2] ✉, Shevaldova OV[1], Kovaleva AV[1]

[1] Federal Research Center for Innovator and Emerging Biomedical and Pharmaceutical Technologies, Moscow, Russia

[2] Dmitry Rogachev National Medical Research Center of Pediatric Hematology, Oncology and Immunology, Moscow, Russia

The psychological effects of childhood cancer and its treatment extend to all family members, particularly healthy siblings, of the affected child. Previous studies have reported reduced quality of life and self-esteem, as well as increased death anxiety and loneliness, among these children. This study aimed to compare the severity of chronic fatigue in siblings of children with cancer and in children from families without a seriously ill family member, with consideration of the informant: the child's self-report or the parent's proxy report. A total of 113 parent–child dyads participated in this controlled cross-sectional study. The sibling group comprised 65 healthy children whose brother or sister had cancer, and the control group comprised 48 children from families without a seriously ill family member. The groups were matched for the children's sex and age (7–11 years). Chronic fatigue was assessed using the Chronic Fatigue Severity Questionnaire. Linear mixed-effects models revealed a significant group × informant interaction ($p < 0.001$). In the control group, parents rated fatigue severity lower ($\Delta = -10$ points) than did the children themselves. In the sibling group, parental ratings were comparable to or higher than the children's self-ratings ($\Delta = 2$ points, $p < 0.001$). Parent–child agreement was stronger in the sibling group ($r = 0.73$, $p < 0.001$) than in the control group ($r = 0.34$, $p = 0.019$). No between-group differences were found in the total fatigue scores based on the children's self-reports. Siblings reported greater physiological discomfort ($p = 0.037$), but less decline in general well-being, cognitive comfort, and social functioning than controls (all $p < 0.001$). The results suggest that in families of children with cancer, parental assessments of a healthy child's fatigue differ from the child's own perception of their condition. Parents tend to rate fatigue as more severe than do the children themselves, whereas the opposite pattern is observed in control families. This informant discrepancy should be considered when assessing the well-being of healthy siblings and when providing psychological support to families affected by childhood cancer.

Keywords: chronic fatigue, siblings, parents, family stress, pediatric cancer, rehabilitation

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✉ **Correspondence should be addressed:** Elena N. Likhomanova
Baltijskaya, 8, Moscow, 125315; likhomanova_en@academpharm.ru

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ХРОНИЧЕСКОЕ УТОМЛЕНИЕ У СИБЛИНГОВ ДЕТЕЙ С ОНКОЛОГИЧЕСКИМИ ЗАБОЛЕВАНИЯМИ: САМООТЧЕТЫ ДЕТЕЙ И НАБЛЮДЕНИЯ РОДИТЕЛЕЙ

Е. Н. Лихоманова[1,2] ✉, О. В. Шевалдова[1], А. В. Ковалева[1]

[1] Федеральный исследовательский центр оригинальных и перспективных биомедицинских и фармацевтических технологий, Москва, Россия

[2] Национальный медицинский исследовательский центр детской гематологии, онкологии и иммунологии имени Д. Рогачева, Москва, Россия

Психологические последствия лечения онкологического заболевания ребенка затрагивают всех членов семьи, в частности здоровых братьев и сестер (сиблингов). Известно о снижении качества их жизни, самооценки, страхе смерти и одиночества. Цель исследования — сравнить уровень хронического утомления у сиблингов детей с онкологическими заболеваниями и у детей из семей без тяжелобольных близких в зависимости от информанта: по данным самооценки ребенка и наблюдения родителя. В поперечном контролируемом исследовании заполняли опросник «Степень хронического утомления» 113 диад «родитель–ребенок»: 65 пар, где есть второй больной ребенок (группа сиблингов), и 48 контрольных пар. Группы были сопоставимы по полу и возрасту детей (7–11 лет). Линейными смешанными моделями выявлено значимое взаимодействие «группа × информант» ($p < 0{,}001$). В контроле родители занижали уровень утомления в сравнении с самооценкой детей (–10 балла), а у сиблингов родительские оценки были сопоставимы с детскими или превышали их по большинству шкал, кроме социальной сферы ($\Delta = 2$ балла, $p < 0{,}001$). Внутренняя корреляция в группе сиблингов оказалась сильнее ($r = 0{,}73$, $p < 0{,}001$), чем в контрольной ($r = 0{,}34$, $p = 0{,}019$). Межгрупповые самооценки детей по общему баллу не различались, но у сиблингов выраженность физиологического дискомфорта была выше ($p = 0{,}037$), а снижение общего самочувствия, когнитивного дискомфорта и нарушения в социальной сфере — ниже ($p < 0{,}001$). Вывод: в семьях с тяжелобольным ребенком родители систематически завышают уровень утомления здорового ребенка, а сами дети оценивают свое состояние иначе. Это расхождение необходимо учитывать при медико-психологическом сопровождении семей.

Ключевые слова: хроническое утомление, сиблинги, родители, семейный стресс, детская онкология, реабилитация

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✉ **Для корреспонденции:** Елена Николаевна Лихоманова
ул. Балтийская, д. 8, г. Москва, 125315; likhomanova_en@academpharm.ru

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Malignant neoplasms (MNs) are among the leading causes of mortality in children. In Russia, in 2024, MNs were reported in 0.013% of the population aged 0–17 years (12.7 cases per 100,000 population); of these cases, 32.4% were stage I–II MNs, and the incidence peaked in children under 4 years of age [1]. In 2022, the global incidence exceeded 211,000 cases per year [2]. Russian population-based studies have reported a steady increase in the survival rate of children with cancer, from 82.4% in 2000 to 89.0% in 2021 [3], reaching 95% for certain types of cancer [4]. In this context, the focus of researchers and clinicians has shifted to the future quality of life of both patients and their families [5].

The consequences of childhood cancer affect every family member. The family's usual way of life and distribution of responsibilities change, and family income often decreases [6–9]. In this situation, healthy brothers and sisters of the affected child (siblings) require particular attention. Their difficulties often go unnoticed: parents who are focused on the affected child may be unable to adequately monitor the well-being of their other children, while clinicians usually do not work directly with them [10, 11]. Although the mean anxiety and depression scores of most siblings remain within age-appropriate ranges, many experience symptoms of post-traumatic stress, reduced quality of life, and problems at school [10]. According to studies conducted by Russian-speaking authors, low self-esteem, fear of death, loneliness, auto-aggression, and anxiety are common among siblings [12, 13]. With regard to somatic problems, siblings have been reported to have higher rates of pain complaints, sleep and eating problems [14], and emergency department visits [15], which may reflect allostatic load, that is, persistent physiological alterations caused by chronic stress [16]. A study involving a large cohort of families with seriously ill children showed that healthy members of such families sought medical care significantly more often, regardless of sex [5].

The feeling of persistently increased fatigue is one of the common manifestations of accumulated stress [5, 15]. In this paper, fatigue is considered not as a clinical diagnosis, but as a subjective feeling of reduced energy — a persistent feeling that children are able to describe themselves. Fatigue can affect physical well-being, emotional state, cognitive functioning, and social motivation. The reliability of such self-assessment in school-age children (from approximately 7–8 years of age) has been confirmed in numerous studies [17, 18].

Parents' proxy reports (parents' perceptions of their children's condition) are widely used in clinical practice and scientific research alongside children's self-reports. Comparing these two sources of information represents a separate scientific issue. Studies involving children undergoing cancer treatment have shown that parents tend to systematically rate their children's fatigue as more severe than the children do themselves. In particular, a multicenter study conducted in 2020 found that parents rated fatigue, on average, 13.7 points higher than the children themselves, and the parents' own fatigue severity further increased this discrepancy [17]. A questionnaire for assessing fatigue in children with cancer aged 7–17 years has been translated into Russian and validated [19]. The questionnaire includes two versions: a child self-report and a parent proxy report based on the parent's observations. The question of how parent and child assessments compare in families in which a child with cancer has healthy siblings has received little attention in the scientific literature. However, parents experiencing chronic stress and primarily focused on caring for a child with cancer and monitoring his or her physical condition may also be particularly sensitive to physical symptoms in their healthy children.

This study focused on assessing chronic fatigue in siblings of children with cancer compared with healthy children who did not have a seriously ill brother or sister. The analysis was based on two sources of information (the child's self-report and the parent's proxy report) and examined the nature and direction of discrepancies between these sources in the two groups. The research hypothesis was that the severity of chronic fatigue in siblings of children with cancer differs significantly from that in children from families without a seriously ill family member, and that the nature of these differences depends on the source of information (child self-report vs. parent proxy report). The study aimed to compare the severity of chronic fatigue in siblings of children with cancer and in children from families without a seriously ill family member, taking into account the informant (child self-report vs. parent proxy report).

METHODS

The study included 113 parent–child dyads divided into two groups. Only mothers played the role of "parents". The group of siblings of children with cancer (hereinafter "siblings") consisted of 65 dyads, in whose families the followed-up children with cancer were raised. Among siblings, relative to the cancer survivors, there were four fraternal twins, 29 younger and 31 elder siblings. The sex was concordant in 32 sibling pairs and discordant in 33 pairs. The median age difference with the survivors was 3.11 years [IQR: 3.10; 4.48]. The maximum number of children per family in this group was 8. In the group of siblings, 24 families (37%) were multi-child (three or more children). Inclusion criteria for siblings: living in the same household as the affected child before the cancer diagnosis and for at least one year thereafter; no history of acute ot chronic disorder, no acute ot chronic disorder at the time of assessment (including the period of 14 days before the beginning of the study); taking no medications; fluency in Russian.

The control group consisted of 48 dyads in which the assessed child: was healthy; used no drugs; was fluent in Russian; had no close relatives with life-threatening disorders. Among them there were 18 elder, three middle, 10 younger, and 17 only children (there were no twins). Parents of the control group had a maximum of three children; there were only five multi-child families (12.5%), and in three of those (60%) the eldest children were full-aged and lived apart from the family.

Groups were matched by age (Me = 9.65 [IQR: 8.36; 11.0] years in the control group and Me = 9.56 [IQR: 8.59; 10.9] years in the group of siblings; $p = 0.454$), gender (50.0 and 52.3% of girls, respectively; $p = 0.788$). Groups were not matched by the fact of having/not having many children (Fischer's exact test; $p = 0.005$).

The standardized Chronic Fatique Severity questionnaire was used to assess the chronic fatigue severity subjective perception [20]. The method consists of 36 items, each of which is graded on a three-point scale (unambiguous agreement, ambiguous agreement or disagreement). The total score represents the Chronic Fatigue Index (CFI) allowing one to estimate the condition severity: no signs of fatigue, early stage, advanced stage, severe chronic fatigue and asthenic syndrome (transformation into disease).

In addition to CFI, the questionnaire includes another four subscales characterizing manifestations of chronic fatigue in various spheres of life. Since the number of points per subscale is unequal, the authors recommend convert these into percentages to ensure the correct interscale comparison. The original version of the questionnaire was designed for assessment of adults, so it was adapted for children: lexemes with the root "work" were replaced by lexical equivalents of

Table 1. Chronic Fatique Severity questionnaire subscale and authorized abbreviations

Subscale full title	Short title used in the text	Reliability indicators
Physiological Discomfort Symptoms	Physiological Discomfort	α = 0.756 ω = 0.786
General Well-Being Decline and Cognitive Discomfort	General Well-Being Decline	α = 0.710 ω = 0.726
Emotional and Affective Disturbances	Emotional Disturbances	α = 0.813 ω = 0.817
Decreased Motivation and Social Communication Changes	Social Functioning Impairment	α = 0.738 ω = 0.742
Total score (chronic fatigue index, CFI)	Total score / CFI	α = 0.847 ω = 0.848

educational activities: "learning" (No. 12, 28), "learn" (No. 21, 25), and "school" (No. 33). The internal consistency evaluation (Cronbach's alpha (α) and McDonald's omega (ω)) confirmed the modified method validity (according to the generally accepted threshold α, ω ≥ 0.7 [21]). Short titles of the subscales used below and appropriate reliability values are provided in Table 1.

Testing was performed in Russian and implemented on an online platform. Data from mothers were acquired remotely: they filled the questionnaire at a place and time convenient for them. Children completed the questionnaire in person in the presence of a clinical psychologist on a provided computer. The child completed the questionnaire either independently or in the form of a semi-structured interview, in which a specialist read the questions aloud, helped to interprete instructions, and recorded the child's answer options. Thus, two independent estimates of the condition of each child were obtained for each dyad: self-assessment and parent's assessment. Furthermore, subjects answered the survey questions about the frequency of getting ill (ARVI) throughout the year, as well as about regular sports, music, and dancing.

Statistical data processing was performed in Jamovi version 2.3.28 (The jamovi project, Australia). Quantitative variables were described using the median (Me) and interquartile range [Q_1; Q_3]; categorical variables were described using absolute and relative frequencies (*n*, %). The Mann–Whitney U-test was used to compare two independent groups based on quantitative indicators; the Kruskal–Wallis test was used for three or more independent groups; Pearson's chi-squared test (χ^2) or Fishers'exact test (with the expected frequencies less than 5) was used for categorical variables.

Linear Mixed Models (LMM) with the parameter estimation by the Restricted Maximum Likelihood (REML) method were used as the main analysis method. The nested data structure was considered by including a random participant effect (random intercept based on the dyad ID), which allowed us to correctly model the relationship between the child's and parent's assessment within the same dyad. The following were included in the model as fixed effects: group (control/siblings), informant (child/parent), and their interaction (group × informant). Covariates included the child's sex, frequency of acute respiratory disorders, and regular exercise (sports and dancing).

The parents' and children's assessment coherence was determined using two additional methods: we calculated the Spearman's rank correlation coefficient for paired scores within each group; the difference Δ = (parent's assessment) — (child's assessment) was also calculated for each dyad; groups were compared based on the Δ values using the Mann–Whitney U-test. The Wilcoxon test for related samples was used to assess intergroup differences between the parents' and children's estimates. The critical significance level for all analyses was as follows: $p < 0.05$.

RESULTS

There were no age and sex differences between the control group and the group of siblings. ARVI was significantly more often reported in the group of siblings (more than five episodes per year): 41.5% vs. 20.8% in the control group ($p = 0.001$). Siblings were less often engaged in sports (35.4% vs. 56.3%; $p = 0.002$) and dancing (16.9% vs. 31.3%; $p = 0.016$) (Table 2). None of the fatigue indicators turned out to be associated with the birth order — neither in the group of siblings ($p > 0.4$), nor in the control group (Kruskal–Wallis test; $p > 0.2$). The age difference (fraction of a year) between the healthy and affected siblings turned out to have no significant association with any of fatigue indicators (Spearman's rank correlation coefficient; $p > 0.05$). In the group of siblings, when the healthy sibling's sex did not match the affected one's sex, the CFI was higher according to both chilren's self-reports ($U = 270$, $p = 0.003$; $Me_{same\ sex} = 13.0$, $Me_{different\ sex} = 25.0$) and mothers' proxy-reports ($U = 307$, $p = 0.015$; $Me_{same\ sex} = 17.0$, $Me_{different\ sex} = 24.0$). Furthermore, the children's self-assessment was higher in siblings of different gender based on two subcales: Physiological Discomfort ($U = 201$, $p = 0.019$; $Me_{same\ sex}$ =

Table 2. Socio-demographic and clinical and anamnestic characteristics of the sample

Indicator	Control (*n* = 48)	Siblings (*n* = 65)	*p*
Age, Me [Q_1–Q_3], years	9.65 [8.36–11.0]	9.56 [8.59–10.9]	0.454
Girls, *n* (%)	24 (50.0%)	34 (52.3%)	0.788
Frequent ARVI (>5 episodes/year), *n* (%)	10 (20.8%)	27 (41.5%)	0.001
Regular sports, *n* (%)	27 (56.3%)	23 (35.4%)	0.002
Regular music, *n* (%)	9 (18.8%)	14 (21.5%)	0.738
Regular dancing, *n* (%)	15 (31.3%)	11 (16.9%)	0.016

Note: The data are presented as Me [Q_1–Q_3] or *n* (%). Quantitative variables were compared using the Mann–Whitney U-test; categorical ones were compared using Pearson's chi-squared test (χ^2) or Fischer's exact test.

Table 3. Chronic fatigue indicators by subscales and linear mixed model results

Subscale	Group	Parents, Me [Q_1–Q_3]	Children, Me [Q_1–Q_3]	*p* (group)	*p* (informant)	*p* (group × informant)
Total score (CFI)	Control	12.0 [6.0–16.0]	20.0 [16.0–25.0]	0.002	< 0.001	< 0.001
	Siblings	22.0 [15.0–29.0]	20.0 [12.0–28.0]			
Physiological Discomfort	Control	7.0 [0.0–10.0]%	13.0 [7.0–20.8]%	< 0.001	0.002	0.005
	Siblings	23.0 [10.0–30.0]%	20.0 [13.0–30.0]%			
General Well-Being Decline	Control	14.1 [0.0–26.3]%	30.0 [20.0–40.0]%	0.513	0.009	< 0.001
	Siblings	30.0 [15.0–40.0]%	20.0 [10.0–30.0]%			
Emotional Disturbances	Control	17.0 [0.0–33.0]%	25.0 [17.0–42.0]%	0.01	0.508	< 0.001
	Siblings	49.4 [25.0–66.7]%	25.0 [10.0–58.3]%			
Social Functioning Impairment	Control	40.0 [10.0–60.0]%	60.0 [50.0–70.0]%	0.58	< 0.001	< 0.001
	Siblings	40.0 [30.0–60.0]%	50.0 [30.0–60.0]%			

Note: The data are presented as the median and interquartile range [Q_1–Q_3]; for subscales — as the percentage of the maximum score. LMM with fixed effects of the group, informant, and their interaction; random effect — dyad ID. Covariates: frequency of ARVI, engagement in sports and dancing.

13.0, $Me_{different\ sex}$ = 28.5) and Social Functioning Impairment (U = 205, p = 0.023; $Me_{same\ sex}$ = 40.0, $Me_{different\ sex}$ = 60.0). The number of children in families with affected children was correlated to none of the fatigue parameters (p > 0.17 in all cases).

Comparison of groups based on the chronic fatigue severity: linear mixed model results

LMMs were used to assess intergroup differences considering the "dyad" data structure. The CFI analysis revealed significant effects of the group (p = 0.002), informant (p < 0.001), and their interaction (p < 0.001). This suggests that the nature of differences between the parents' and children's estimates is significantly different in two groups (Table 3).

In the control group, parents regularly underestimated the child's fatigue severity compared to the children's self-assessment (Me_{CFI}: 12.0 [6.0–16.0] in parents vs. 20.0 [16.0–25.0] in children). In the group of siblings, the pattern was inverted: the parents' estimates were comparable with the children's or higher (22.0 [15.0–29.0] in parents vs. 20.0 [12.0–28.0] in children).

The analysis of subscales using the LMM showed that the "group × informant" interaction was significant for all the studied indicators (all p < 0.01), which suggests the systemic nature of the differences revealed (Table 3).

The effect of the group was most pronounced for the subscales Physiological Discomfort (p < 0.001) and Emotional Disturbances (p = 0.010): the siblings' parents assessed the children's condition as more severe, than parents of the control group, based on both indicators. As for the General Well-Being Decline (p = 0.513) and Social Functioning Impairment (p = 0.580) subscales, the effect of the group was non-significant, but the significant interaction was preserved, which suggests different patterns of estimates in the groups. In most cases, the covariates included had no significant effect on the model results.

Coherence of parents' and children's estimates

To thoroughly assess the nature of the "group × informant" interaction, we conducted a number of additional analyses involving assessment of correlations within groups, analysis of the difference Δ, and pairwise comparison of estimates within the dyads.

The correlation analysis (Table 4, Fig. 1) revealed significant differences in coherence of estimates between groups. In the control group, the correlation between the parents' and children's CFI assessment was weak (r = 0.34, p = 0.019). A strong positive correlation was observed in the group of siblings (r = 0.73, p < 0.001), which suggests much higher coherence of the child's condition perception in the dyads of siblings.

The analysis of the difference Δ confirmed a fundamental difference in the nature of the ratio of estimates. In the control group, parents underestimated the children's fatigue: MeΔ = −10.0 [−14.0; −3.0] points. In the group of siblings, the estimates were comparable or slightly exceeded the chidlren's self-assessment (MeΔ = 2.0 [−4.0; 4.0]). The intergroup differences in Δ values were significant (Mann–Whitney U-test: U = 625, p < 0.001, r = 0.6).

Intragroup and intergroup differences in parents' and children's estimates

We conducted a pairwise comparative analysis of parents' and children's estimates both within each group and between groups. The results are provided in Fig. 2.

In the control group, the parents' estimates were significantly lower, than the children's, based on all subscales and CFI (p ≤ 0.045), which suggests a persistent tendency among parents of healthy children to underestimate the fatigue severity in their children. In the group of siblings, the pattern was inverted: there were no differences between the parents' and children's estimates based on the Physiological Discomfort

Table 4. Correlation between the parents' and children's assessment of chronic fatigue severity (CFI) by groups

Group	Spearman's *r*	*p*
Control (n = 48)	0.34	0.019
Siblings (n = 65)	0.73	< 0.001

Note: Spearman's rank correlation coefficients between the parent's and child's estimates within the same dyad are provided.

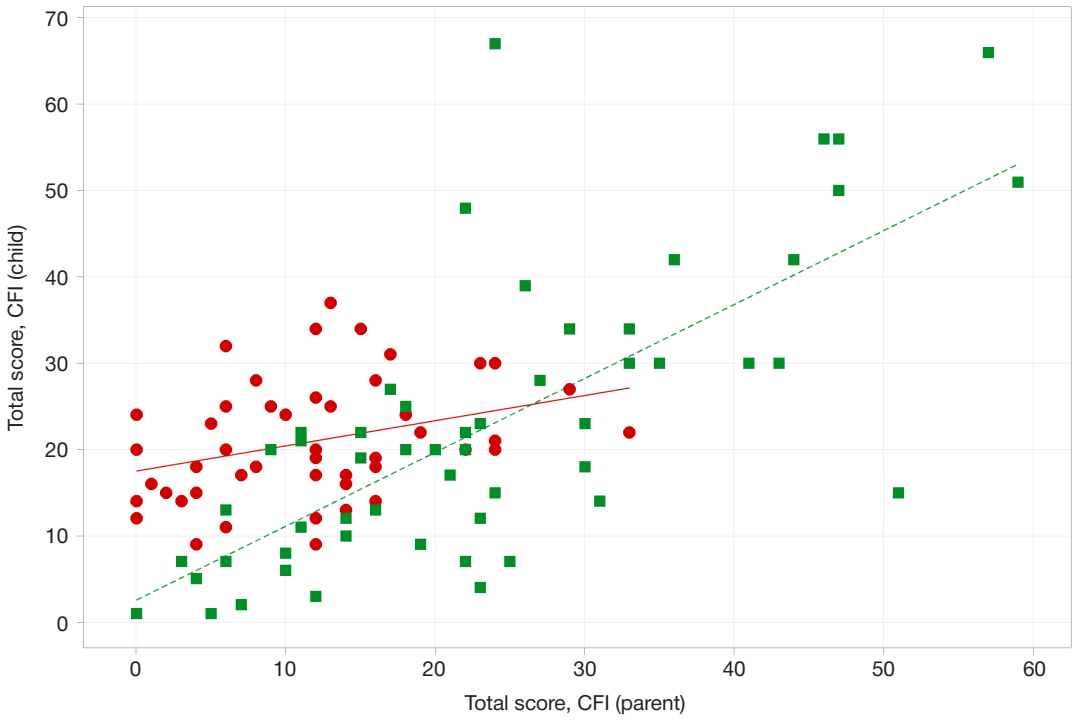

Fig. 1. Scatter charts with the linear regression lines for CFI assessment by parents and children in the control group (*red*) and siblings (*green*). X axis — parent's estimate; Y axis — child's estimate.

and Social Functioning Impairment subscales, as well as total CFI ($p = 0.997$; $p = 0.474$; $p = 0.241$, respectively). Significant differences directed towards overestimation by parents were reported for the General Well-Being Decline ($p = 0.005$) and Emotional Disturbances ($p = 0{,}002$) subscales.

When performing an intergroup comparison separately by informants, the pattern also turned out to be asymmetric. According to the parents, significant intergroup differences were revealed based on four indicators out of five: Physiological Discomfort, General Well-Being Decline, Emotional Disturbances, and total CFI (all $p < 0.001$). According to the children's self-reports, significant differences were reported only based on distinct subscales: Physiological Discomfort ($p = 0.037$), General Well-Being Decline ($p < 0.001$), and Social Functioning Impairment ($p = 0.016$). There were no significant differences in children's self-assessment based on the total CFI and the Emotional Disturbances subscale ($p = 0.729$ and $p = 0.631$, respectively). Thus, the parents', not children's, estimates were the main contributors to intergroup differences, which is consistent with the linear mixed model results.

DISCUSSION

There were significantly more multi-child ones among the families, where children with cancer were raised, which is in line with the large sociological study of 1298 Russian parents having children with cancer [12]. It was demonstrated, how the existential stress faced by such families was associated with changes in their life purpose orientations and the system of needs, which, taken together, resulted in the multi-child lifestyle.

The sex difference rate in both groups turned out to be associated with the chronic fatigue severity: it was higher in cases where the siblings were of different sexes. This effect was revealed when performing further analysis, so we can only put forward a number of hypotheses discussed in the literature and requiring further research. Parents may behave differently with children of different sexes, making different demands and expectations on them, building emotional relationships with them in different ways, devoting time to them in different proportions [22]. It should be noted that the literature data are controversial [23–25]. According to large family-based studies of behavioral and psychological traits, the same genes contribute to interindividual differences in both males and females, which makes the genetic explanation of the sex differences identified less likely compared to socio-psychological ones [26].

Intergroup differences in the children's lifestyle and health revealed that siblings in our sample were less likely to engaged in sports and dancing, twice more often got ARVI (Table 2). These findings are consistent with the results of different studies showing that brothers and sisters of childhood cancer survivors more often seek medical care compared to healthy peers [5, 14, 27]. The siblings' lower engagement in the organized physical activities is natural: the family schedule restructuring, deterioration of the family's financial resources, parents' focus on the sick child's needs, and the sibling's own anxiety reduce the time and resources for leisure activities [10, 28]. Considering the fact that regular physical activity represents one of the reliably determined factors protecting children against fatigue [29], the physical activity decrease can be an intermediate between chronic family stress and subjective feeling of fatigue. At the same time, motor activity was included in the linear mixed models as a covariate, but it did not explain the main effects, which suggests that there were additional mechanisms.

As for CFI, there were no significant differences in children's self-assessment in two groups, but the analysis of subscales revealed a more differentiated pattern. The siblings' Physiological Discomfort subscale scores were higher, than that of the control group, while differences on the General Well-Being Decline subscale were oppositely directed: the scores of control children turned out to be significantly higher. There were no significant differences in self-assessment of two groups on the Emotional Disturbances subscale, while the Social Functioning Impairment subscale scores of control children were also significantly higher. Thus, according to the children's self-assessment, the fatigue indicators in siblings are significantly higher compared to that in the control group based

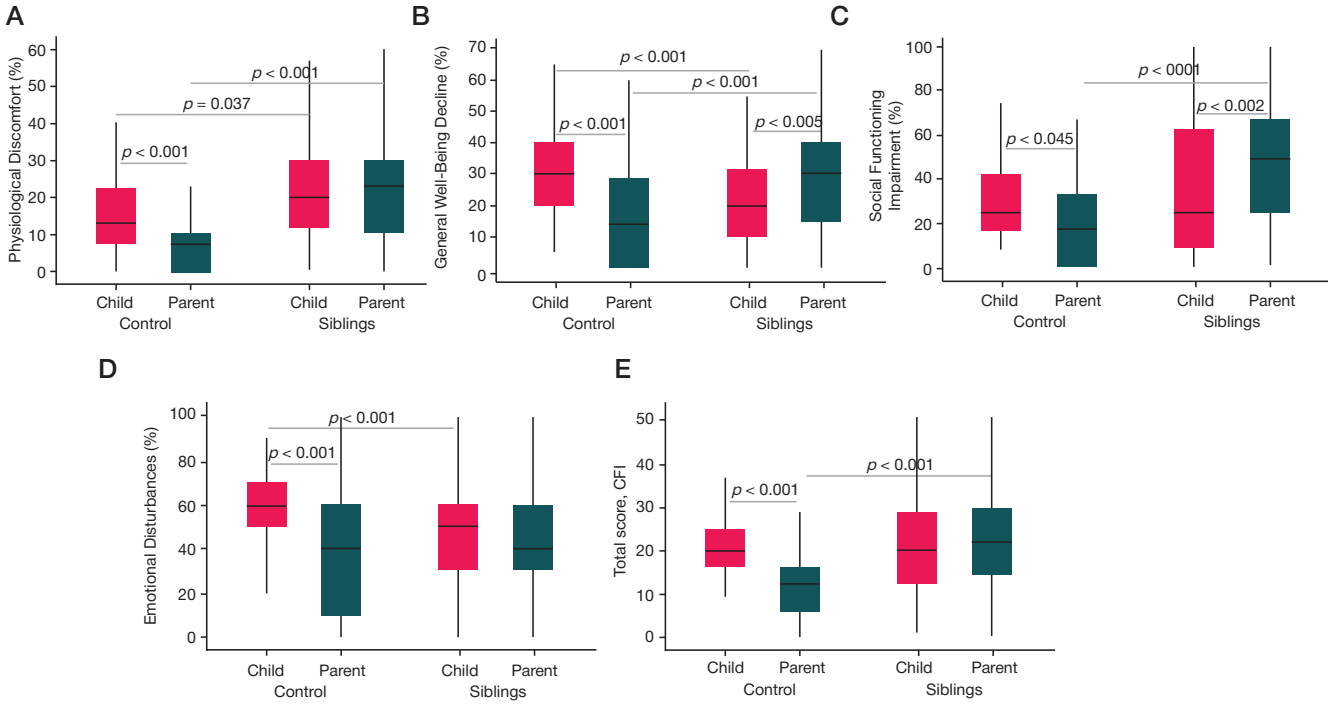

Fig. 2. Box plots of the distribution of chronic fatigue indicators by subscales in the control group and group of siblings. *Red box plots* — children's estimates; *green* — parents' estimates. Intragroup differences between the child's and parent's estimates (Wilcoxon test for related samples) and intergroup differences between the control group and group of siblings (Mann–Whitney U-test) are designated by red brackets with *p*-values. Only statistically significant comparisons are shown ($p < 0.05$). **A**. Physiological Discomfort Symptoms subscale. **B**. General Well-Being Decline and Cognitive Discomfort subscale. **C**. Emotional and Affective Disturbances subscale. **D**. Decreased Motivation and Social Communication Changes subscale. **E**. chronic fatigue index (CFI, total score)

on the physiological component only; there are no differences or indicators of the control group are significantly higher in the emotional, cognitive-affective, and social spheres. The latter can reflect the acceleration effect of the chronic family stress load: siblings of childhood cancer survivors often show higher levels of pro-social behavior and empathy, as well as a tendency not to transmit their own difficulties, feeling an unspoken prohibition against demonstrating weakness in a situation of family stress [10]. Parents more often ask healthy siblings for instrumental help (for example, more often ask for help with their chores), despite the fact that parents themselves less often provide similar support to their children (for example, less often allow them to skip school) [11]. Furthermore, acceleration processes were illustrated by the siblings' anthropometric measurements: their body height and body mass index were higher compared to that of their peers (according to the WHO), and the higher body height was reported in siblings, who were older, than survivors [30].

When comparing the parents' and children's estimates within each group, it was found that in the control group parents rated the child's fatigue lower, than children, on all five scales. This is consistent with the data showing that the parents' assessment based on their insights is often lower compared to the children's self-assessment regarding subjective, especially emotional and social, aspects of the condition, since the age of 7 [31]. The pattern was inverted in the group of siblings. The parents' General Well-Being Decline and Emotional Disturbances subscale scores were significantly higher compared to children's. As for Physiological Discomfort, Social Functioning Impairment, and total CFI, there were no differences between the parents' and children's estimates in the group of siblings.

At the age of 7–11 years children actively shape their internal pictures of illness (IPI) and health (IPH). At this age, subjective well-being is already accessible to reflection, but the assessment of one's condition as health or illness still depends heavily on the interpretations and reactions of significant adults [32]. The developmental trajectory of the IPI/IPH involves a gradual shift toward greater autonomy in self-assessment. This pattern was observed in our control group, where children's self-assessments on several subscales exceeded those of their parents. Such a discrepancy suggests that the child has his/her own well-being assessment criteria, that are different from the parents', which reflects the developing ability to rely on the internal sensations and ideas, not only on the ones transmitted by external sources. In the context of chronic family stress experienced by the family having a child with the life-threatening disease, the typical development can be modified. The constant presence of a child with the objectified suffering in the family and the transmitted parents' anxiety can "shift" the criteria of normal [33]. Thus, the lack of significant differences between the parents' and children's estimates in the group of siblings can reflect not so much the objective differences in the children condition, but rather the features of the sibling's IPI/H ontogenesis, during which his/her own IPI/H remains "fused" with parental assessments and subject to the anxiety of the family environment for longer.

The linear mixed model analysis revealed a significant interaction of the "group × informant" factors for all the studied indicators. Intergroup differences are ensured mainly by the parents', not children's, estimates. According to the parents, fatigue indicators on all subscales, except the social one, are higher in the group of siblings. The pattern based on the children's self-assessment is different: based on the General Well-Being Decline and Social Functioning Impairment siblings described their condition as better compared to control children, while based on Physiological Discomfort the condition was described as slightly worse. Such an asymmetric pattern, with which intergroup differences are determined primarily by the parents' estimates, is consistent with the hypothesis about the indirect influence of parents' assessment on the IPI shaping in siblings of children with cancer.

In 2020, it was shown that the parents' awareness about the sibling's conditions and accuracy of their assessment are determined primarily by the quality of communication in the family. Chronic stress associated with the other child's illness, being overwhelmed with care and focused on the cancer patient's needs limit the parents' capability of monitoring the healthy child's condition and discuss his/her experiences with him/her [34]. In the context of family crisis, parents often develop the increased anxiety about the children's health in general — the phenomenon described within the framework of the concept of the "child's health anxiety by proxy": excessive, difficult to control fears about the child's possible ill-being, which are not necessarily consistent with the child's actual condition [33]. Parents of children with cancer often experience significant fatigue and emotional distress themselves [35]. The caregiver's own health status and well-being are associated with the discrepancy between his/her and child's estimates. In particular, such discrepancy increased as the caregiver's own condition worsened based on the fatigue domain [14]. A combination of these factors (low awareness resulting from the lack of communication, background anxiety for a healthy child, the parent's own distress) creates conditions for inflated estimates of the sibling's condition.

Coherence of parents' and children's estimates in the group of siblings was significantly higher, than in the control group. Similar results were obtained by the researchers, who showed that coherence of parental and child reports was higher in families affected with cancer. Apparently, under these conditions parents are more sensitive to any changes in children's condition [36]. It must be emphasized that the high correlation reflects the coherence of the direction of changes only, not the accuracy of absolute estimates. In our study, the high correlation was combined with the parents' overestimation on a number of subscales, which underscores the need to differentiate these two aspects when analyzing the inter-informant consensus.

Limitations of the study

The cross-sectional study design made it impossible to examine changes within parent–child dyads over time.

Only subjective measures were collected and analyzed (children's self-reports and mothers' proxy reports); therefore, the influence of social desirability bias cannot be ruled out.

The analysis of the family context was limited. We had no information on the mothers' age at the birth of their first child, their mental health status, or the availability and quality of parenting support. We also lacked information on the degree of fathers' involvement in child-rearing and did not consider the family's socioeconomic characteristics (place of residence, housing conditions, household income, parental education, and employment).

We did not stratify the sibling group according to the clinical characteristics of the child with cancer (cancer diagnosis, treatment duration and intensity, disease recurrence, follow-up period, comorbidities, or symptom severity).

CONCLUSIONS

The study has shown that subjective chronic fatigue in siblings of children with cancer is not a homogeneous phenomenon and is perceived differently by different informants (the child and the parent). According to the children's self-reports, between-group differences are limited to the physiological domain. In contrast, parent proxy reports indicate differences across most of the studied domains and consistently yield higher fatigue ratings than the children's self-reports.

The key finding is the reversal of the discrepancy between parent and child ratings depending on the family context: whereas parents in the control group tend to underestimate their children's fatigue, parents of children with cancer tend to overestimate the fatigue of their healthy children. This discrepancy itself represents an important indicator that should be taken into account when conducting psychosocial screening of siblings.

These findings support the inclusion of healthy siblings in psychological follow-up programs for families in pediatric oncology and rehabilitation. Further longitudinal studies incorporating parental stress and anxiety as predictors of parent–child agreement will help clarify the mechanisms underlying the observed phenomenon and facilitate the development of more accurate recommendations for supporting families affected by childhood cancer. In the present study, we did not consider several potentially important factors, including family composition, socioeconomic status, the availability and nature of social support, and attachment style; these factors should be addressed in future research.

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