

Original Article

Quality-of-Life Evaluation of Healthy Siblings of Children with Chronic Illness

Dinleyici et al. Health-Related QoL in Healthy Siblings

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Background: Chronic illness during childhood can also cause changes in the health-related quality of life (HrQoL) of the individual's family members.

Aim: This study aimed to evaluate the HrQoL among healthy siblings of children with chronic illness.

Study design: Cross-sectional study

Methods: We enrolled healthy siblings of 191 children with a chronic illness (cerebral palsy, epilepsy, diabetes, celiac disease, hematological oncologic disease, or asthma) and those siblings of 100 healthy children for quality of life evaluation. We administered the Pediatric Quality of Life Inventory questionnaire; the physical health and psychosocial health score were calculated using individual sibling and parent responses. The primary endpoint was the comparison of HrQoL scores of healthy siblings of 191 children with a chronic illness and those siblings of 100 healthy children.

Results: The physical health, psychosocial health, and total health scores of healthy siblings of children with a chronic illness were significantly lower than those of siblings of healthy children ($p < 0.001$). In the chronic disease group, the lowest psychosocial health score was found in the cerebral palsy, hematologic/oncologic disease and asthma groups ($p < .001$). The global impact on the quality of life for healthy siblings of children with a chronic disease was significantly higher in children's self-reports than in parents' reports (30.4% vs. 15.1%, $p < 0.05$).

Conclusion: This study showed that most of the healthy siblings of children with a chronic illness are physically and psychosocially affected. In healthy siblings, we observed a global impact on the HrQoL, including psychosocial scores, and a low level of parental awareness about this situation. This might increase the risk of emotional neglect and abuse in these children. Thus, special support programs are needed for the families of children with chronic illness.

Key words: quality of life, children, sibling, chronic illness

A chronic illness is defined as a long-term, untreatable condition or current symptoms that limit activities of daily living (Halfon and Newacheck, 2010). The diagnosis of a physical chronic condition during childhood is a source of serious stress in the family that may lead to changes in the family structure and result in labor loss (Rodrigues and Patterson, 2007). It also has psychosocial impacts on the siblings of the children with a chronic

illness (Barlow and Ellard, 2006). The development of depression and anxiety was more common in the siblings of children with a chronic illness, and they also had less communication with their peers. have an increased risk of internalizing problems and a negative self-concept (Barlow and Ellard, 2006; Limbers and Skipper, 2014). The Pediatric Quality of Life Inventory (PedsQL), a tool for measuring the HrQoL in children, and the PedsQL 4.0 Measurement Model is a multidimensional questionnaire consisting of physical, emotional, social, and school functioning domains (Varni et al., 2001). QoL questionnaires can be administered to the sibling of an ill child, to the child himself/herself, or to the parents and/or healthy children (Limbers and Skipper, 2014). As of 2019, six studies have used the PedsQL to evaluate the HrQoL among siblings of children with a chronic illness (Bansal et al., 2014; Hamblion et al., 2011; Norris et al., 2010; Packman et al., 2005; Wood et al., 2008; Havermans et al., 2015). In the studies conducted so far, the number of participants ranged from 10 to 131, with a mean age of 10 to 13 years; the chronic illnesses studied so far include mainly epilepsy, cancer, hematopoietic stem cell transplantation, type 1 diabetes, and cystic fibrosis (Havermans et al., 2015; Limbers and Skipper, 2014;). The studies were conducted in Canada, Belgium, the United States, India, and the United Kingdom. There is no study on the siblings of children with a chronic illness involving multiple patient groups in Turkey (Limbers and Skipper, 2014; Havermans et al., 2015). HrQoL assessments of siblings of children with a chronic illness allow the assessment of increased functional loss in specific subgroups of illnesses and provide guidance on what can be done to reduce any negative impact on the group of siblings at higher risk (Trama and Dieci, 2011).

In this study, we aimed to evaluate the HrQoL scales of siblings of children with a chronic illness and compare them to those of siblings of healthy children. We also aimed to compare the HrQoL scores of groups with different chronic illnesses. Based on the data from the HrQoL assessments among siblings of children with a chronic illness, we aimed to gain insight into preventive and supportive strategies for groups of patients with an impaired QoL.

MATERIALS AND METHODS

This case-control study was carried out in the inpatient and outpatient services of the XXXXXXXXXXXXX,. Our target population was composed of children aged 2 to 18 years with a chronic illness who were followed up by the XXXXXXX and subspecialty inpatient and outpatient services of the XXXXXXXXXXX between 2016 and 2017, as well as their parents and healthy siblings. The sample consisted of 200 children aged 2 to 18 years with a chronic illness—including hematologic/oncologic malignancy, type 1 diabetes, celiac disease, epilepsy, cerebral palsy, and asthma—who availed of the inpatient and outpatient services of the XXXXXXX, between May 2016 and May 2017. We also included 100 healthy children aged 2 to 18 years without any chronic illness, who presented to the outpatient clinics during the same period. The target population also consisted of at least one parent and at least one healthy sibling of these children. Prior to the study, approval was obtained from the Institutional Ethics Committee for Non-Interventional Clinical Trials.

Inclusion Criteria

- 1- Children diagnosed with a chronic illness such as hematologic/oncologic malignancy, type 1 diabetes, celiac disease, epilepsy, cerebral palsy, and asthma at least three months before the study and healthy children aged 2 to 18 years without any chronic condition and need for regular use of medications
- 2- Healthy siblings of children diagnosed with a chronic illness such as hematologic/oncologic malignancy, type 1 diabetes, celiac disease, epilepsy, cerebral palsy, and asthma at least three months before the study and healthy siblings of healthy children aged 2 to 18 years without any chronic condition and need for regular use of medications

Primary and Secondary Endpoints

Primary endpoint: The primary endpoint of the study was to evaluate the HrQoL scores of the healthy siblings of all children with a chronic physical illness and compare them with those of the siblings of healthy children.

Secondary endpoints: (1) to compare the HrQoL scores of the siblings of children with a chronic physical illness according to their specific disease groups and to compare their scores with those of the healthy siblings of healthy children, (2) to evaluate the HrQoL scores of the siblings of children with a chronic physical illness according to their specific disease groups by gender, and (3) to evaluate the HrQoL scores of the siblings of children with a chronic physical illness according to their specific disease groups by age relative to their ill sibling

Data collection started after consent was obtained from the parents and siblings of the children with a chronic illness who met the inclusion criteria. The demographic characteristics of the children with a chronic condition, as well as of their parents and siblings, were recorded on the questionnaire. We administered the PedsQL to all parents and siblings who were at an appropriate age to complete a questionnaire. The validity and reliability of the Turkish translation of the PedSQL inventory have already been established. Written permission to use the tool was granted by James W. Varni, who developed the tool.

Based on the age group of the siblings of children with a chronic illness, one of the following forms was used: Pediatric Quality of Life Child Form (2–4 years old), Pediatric Quality of Life Child Form (5–7 years old), Pediatric Quality of Life Child Form (8–12 years old), and Pediatric Quality of Life Adolescent Form (13–18 years old). The PedsQL 4.0 consists of 23 questions that cover four domains of HrQoL measurement in children. Two separate forms, the child and parent forms, were completed for all children who participated in the study. We used a 3-response and 5-response visual Likert scale for children below 8 years old and for those aged 8–18 years, respectively. The physical health score was based on the mean score for eight questions on physical functioning, with responses presented as 100 (never), 75 (seldom), 50 (sometimes), 25 (often), and 0 (almost always). The psychosocial health score was based on the mean score of three different categories: emotional functioning, social functioning, and school functioning. The total health score was based on the summary score across all four categories of functioning and ranged from 0 to 100, with higher scores indicating a better QoL. The Child and Adolescent Form of the PedsQL evaluates the past one month of the child or adolescent (Varni et al., 1999).

Statistical Analysis

According to our sample size calculation, each groups needs to include at least 84 children (a power of 0.8, on the basis of 4 predictors, and probability level was 0.05). For potential subgroup analysis for children with chronic disease group, we enrolled 2:1 (chronic disease group :control) and we increased 20% for potential dropouts; totaly 200 children for chronic disease and 100 control disease). Statistical analysis of the data was carried out using the Statistical Package for the Social Sciences (Chicago, IL) for Windows 11.5 software program. The normality of the patients' demographic data and PedsQL scores was analyzed with the Shapiro-Wilks test, which showed that the PedsQL scores were normally distributed. The results were summarized as means and standard deviations. Demographic and categorical data were presented as percentages, and the chi-square test and Student *t* test were used for comparisons. Analysis of variance was used to compare QoL scores among chronic conditions. Since the QoL scores were not homogeneously distributed, the Games-Howell test was used to compare subgroups in cases where there was a statistically significant difference between the groups. A *p* value of less than .05 was considered statistically significant.

RESULTS

The study included siblings of 200 children with a chronic illness—including chronic neurological disease, epilepsy, type 1 diabetes, celiac disease, asthma, and hematologic malignancy—who presented to the inpatient and outpatient clinics of the Department of Pediatrics of the XXXXXX. Nine children were excluded because they did not respond to more than 50% of the questions. A total of 191 healthy siblings were evaluated. The control group included 100 healthy children of similar age with a healthy sibling.

Demographic Characteristics

There was no statistically significant difference in the age, educational level, and socioeconomic level of parents between the study and control groups ($p > .05$). When we evaluated the monthly income of our participants, we categorized as low, middle and high according to our country specific data. In chronic disease group, 11% low, 82% middle and 7% high, and in control group, 8% low, 84% middle and 8% high. No statistically significant difference was found in the educational level and monthly income of parents between the chronic illness group and the control group ($p > .05$).

The study included 191 children with a chronic illness (85 boys and 106 girls). The mean age of children with a chronic illness was 116 ± 56 months. The group of children with a chronic illness included 26 children with cerebral palsy, 39 children with hematologic/oncologic disease, 31 children with asthma, 28 children with diabetes, 33 children with celiac disease, and 34 children with epilepsy. The median time to enrollment in the study from the diagnosis of a chronic illness was 36 months. The median time from diagnosis was 72, 24, 36, and 48 months in the group of children with cerebral palsy, hematologic/oncologic disease, and asthma, the epilepsy group, the diabetes group, and the celiac disease group, respectively. The time from diagnosis was greater in the cerebral palsy group than in the epileptic and asthmatic children ($p < .05$ for both), while no difference was found in the duration of illness among other groups ($p > .05$).

The healthy siblings of children with a chronic illness and the healthy siblings of the control group had a similar age and gender distribution ($p > .05$; Table 4.2). No significant difference was found in the age and gender of siblings between the study and control groups. The mean age of 191 healthy siblings in the study group (102 boys and 89 girls) was 119 ± 58 months. Of these siblings, 101 (52.9%) were older than the index case. The median percentage of siblings of the children with a chronic illness who were older than the index case was 61.5%, 35.9%, 45.2%, 42.9%, 45.5%, and 55.9% in the cerebral palsy, hematologic/oncologic disease, asthma, diabetes, celiac disease, and epilepsy groups, respectively, with no statistically significant difference between the groups ($p > .05$) (Table 1).

Self-Reports of Healthy Siblings

The psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness were significantly lower than those of the healthy siblings of healthy children ($p < .001$, $p < .01$, and $p <$

.001, respectively; Table 2). The mean psychosocial health score of the HrQoL scale was 72.6 ± 13.7 among the healthy siblings of children with a chronic illness vs. 82.1 ± 11.6 in the control group ($p < .001$). The mean physical health score was lower in the study group than in the control group (82.6 ± 13.8 vs. 87.2 ± 8.8 , respectively, $p < .01$). The total PedsQL score (psychosocial and physical health scores combined) was also lower among the siblings of children with a chronic illness than in the healthy control group (75.1 ± 11.6 vs. 83.4 ± 10.4 , respectively, $p < .001$; Table 2).

An analysis of the self-reports of healthy siblings of children in the chronic illness subgroups showed that the psychosocial health scores were lowest in the cerebral palsy, hematologic/oncologic disease, and asthma groups (Table 3). While the psychosocial health score was significantly lower in the cerebral palsy, hematologic/oncologic disease, and asthma groups than in the control group ($p < .05$, $p < .001$, and $p < .05$, respectively), there was no such difference in the diabetes, celiac disease, and epilepsy groups ($p > .05$). The psychosocial health score was lower in the cerebral palsy, hematologic/oncologic disease, and asthma groups compared to the celiac disease group ($p < .05$, $p < .001$, and $p < .01$, respectively). Similarly, the psychosocial health score was lower in the cerebral palsy group than in the diabetes group ($p < .01$) (Table 3).

In the healthy siblings' self-reports, the lowest physical health scores were in the hematologic/oncologic disease group; these scores were significantly different from those of the healthy control group ($p < .05$). No difference was found between the siblings of children with asthma, diabetes, celiac disease, epilepsy, and cerebral palsy and the siblings of healthy children; neither was any difference found between the chronic illness groups ($p > .05$). Based on the healthy siblings' self-reports, the lowest total QoL scores were among the healthy siblings of children with hematologic/oncologic disease, asthma, and cerebral palsy; these scores were significantly different from the score of the control group ($p < .001$, $p < .05$, and $p < .05$, respectively). The total score of children with a hematologic/oncologic disease was lower than that of children with diabetes and celiac disease ($p < .01$ and $p < .001$, respectively), while the total score of children with asthma was lower than that of children with celiac disease ($p < .05$; Table 3). When the psychosocial health, physical health, and total scores of the siblings of children with a chronic illness were evaluated (from the perspective of healthy siblings or parents), there was no correlation with the duration of chronic illness in the group of all chronic conditions ($p > .05$) (Table 3).

Parent Reports for Healthy Siblings

The summary of QoL scale scores for the siblings of children with a chronic illness and the control group was higher in the parent reports than in the healthy siblings' self-reports ($p < .05$). The psychosocial health, physical health, and total scores of the siblings of children with a chronic illness were significantly lower than those of the healthy siblings of healthy children ($p < .001$ for all, Table 4). The mean psychosocial health score was 76.8 ± 12.3 for the healthy siblings of children with a chronic illness vs. 88.1 ± 16.6 for the control group ($p < .001$). The mean physical health score was lower in the chronic disease group than in the control group (80.7 ± 15.4 vs. 87.6 ± 8.5 , $p < .001$). The total PedsQL score was also lower among the healthy siblings of children with a chronic illness than in the healthy control group (77.7 ± 11.3 vs. 88.0 ± 6.0 , $p < .001$; Table 4).

Based on the parent reports, the psychosocial health score was lower among the cerebral palsy, hematologic/oncologic disease, asthma, diabetes, celiac disease, and epilepsy groups than among the healthy siblings of healthy children ($p < .01$ for the cerebral palsy and epilepsy groups and $p < .001$ for the other groups; Table 5). No statistically significant difference was found in the psychosocial health score when chronic illnesses were compared to each other ($p > .05$) (Table 5).

In the parent reports of healthy siblings, the physical health score was lower in the hematologic/oncologic disease and diabetes groups than in the healthy control group ($p < .05$), while no difference was found between the control group and the cerebral palsy, asthma, celiac disease, and epilepsy groups ($p > .05$). There was no difference in the physical health score between each of the chronic conditions ($p > .05$). In the parent reports of healthy siblings, the total QoL score was significantly lower among the children with cerebral palsy, hematologic/oncologic diseases, asthma, diabetes, celiac disease, and epilepsy compared to the control group ($p < .01$, cerebral palsy, epilepsy, and celiac disease vs. control; $p < .001$, hematologic/oncologic diseases, asthma, and diabetes vs. control). No statistically significant difference was found in the total QoL score when chronic illnesses were compared to each other ($p > .05$; Table 5). There was no correlation between the psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness (in the responses of healthy siblings and parents) and the duration of the chronic illness ($p > .05$).

Comparison of Psychosocial Health, Physical Health, and Total Health Scores by Gender

The healthy siblings' of children with a chronic illness self-reports showed no significant difference in psychosocial health, physical health, and total health scores between boys and girls ($p > .05$). Similarly, no significant difference was found for gender in the psychosocial health, physical health, and total health scores among children with a chronic illness from the parents' perspective ($p > .05$).

Comparison of Psychosocial Health, Physical Health, and Total Health Scores by Age of Healthy Siblings

Among the healthy siblings of children with a chronic illness, 101 (52.9%) siblings were older than the index case. An analysis of the healthy siblings' self-reports showed no significant difference in the psychosocial health, physical health, and total health scores between the healthy siblings who were older and younger than the ill children ($p > .05$). Similarly, from the parents' perspective, there was no significant difference in the psychosocial health, physical health, and total health scores between the healthy siblings who were older and younger than the ill children ($p > .05$).

Prevalence of Impaired QoL in Healthy Siblings of Children with a Chronic Illness

In their study on HRQoL measurement, Varni *et al.* stated that a child's total self-reported score of less than 71.44 and a parent-report score of less than 67.44 indicated an "impaired quality of life." In the present study, the healthy siblings' self-reports showed that impaired QoL was higher among the healthy siblings of children with a chronic illness than in the control group (30.4% vs. 14.1%, respectively, $p = .005$). An evaluation of parent reports showed no impairment in the QoL of the control group, whereas 15.1% of the siblings of children with a chronic illness had impaired QoL ($p < .001$). The parents reported less impairment than the healthy siblings did (30.4% vs. 15.1%, respectively, $p < .05$), indicating that parental awareness of impaired QoL in healthy children was poor (Table 6). A comparison between the self-reports of healthy siblings and the parent reports of children with a chronic illness showed no significant difference between the physical health scores and total health scores ($p > .05$), whereas the psychosocial health scores of healthy siblings were lower than those of their parents ($p < .05$).

According to the healthy siblings' self-reports, the prevalence of impaired QoL was 42.1%, 51.6%, 45.0%, and 30.7% in the cerebral palsy, hematologic/oncologic disease, asthma, and epilepsy groups, respectively—higher than in the control group (14.1%). The prevalence of impaired QoL was lower among the healthy siblings of children with diabetes (9.9%) and celiac disease (3.3%) than in the control group (Table 7). According to the parent reports, the prevalence of impaired QoL was 26.9%, 10.2%, 25.8%, 14.2%, 3.0%, and 14.7% in the cerebral palsy, hematologic/oncologic disease, asthma, diabetes, celiac disease, and epilepsy groups, respectively, vs. 0% in the control group. The parent reports showed a lower prevalence of impaired QoL compared to their children's self-reports (Table 7).

An analysis of the parent reports of children with a chronic illness by age group showed no statistically significant difference between the psychosocial health, physical health, and total health scores ($p > .05$). The healthy siblings' self-reports showed no significant differences in the psychosocial health score between age groups but a significant difference in the physical and total health scores ($p < .05$), with older age groups having higher scores than the 5–7 age group.

DISCUSSION

This study showed that the QoL is affected in varying degrees in the majority of healthy siblings of children with a chronic illness. The QoL assessments of healthy siblings of 191 children with a chronic illness at a median time of 36 months following the diagnosis showed an impact on the psychosocial health, physical health, and total health scores of these siblings, which were significantly lower than those of the healthy siblings of healthy children. The impact was seen in both parent reports and healthy siblings' self-reports. Several studies have also shown the psychosocial impact observed in both the self-reports and parent reports of the healthy siblings like our study (Waite-Jones and Madill, 2008; Macleod *et al.*, 2003), while other studies have reported no psychosocial outcomes in healthy siblings (Labay and Walco, 2004). Healthy siblings experience emotions such as withdrawal, aggression, depression, anxiety, guilt, and isolation, with poor school performance and low self-esteem. As a result, they may feel lonely, ignored, excluded, neglected, and rejected (Alderfer *et al.*, 2010; O'Brien *et al.*, 2009; Van Riper, 2003; Wilkins and Woodgate, 2005; Murray, 1999; Bellin and Kovacs, 2006). Although the majority of siblings of children with a chronic illness have been negatively affected, most of these impacts are not considered psychopathological but within normal range (Wood *et al.*, 2008; Sharpe and Rossiter, 2002). However, some of the symptoms or findings should be further examined to see if they are psychopathological or a way of coping with the situation.

The factors associated with the psychosocial impact on the healthy siblings of children with a chronic illness include the type and severity of the chronic illness, the time since diagnosis, and the healthy sibling's age, gender, and coping skills (Elissa Lampe Degelman, 2011). A severe and life-threatening illness has a larger impact on healthy siblings' psychological functioning, and their risk of having emotional and behavioral problems is 1.6–2 times greater (Vermaes *et al.*, 2012). The type and severity of chronic illness or disability may cause some effects that will be reflected by the siblings during adulthood (O'Neill and Murray, 2016; Wolfe *et al.*, 2014). An analysis of the healthy siblings' self-reports showed that the groups with cerebral palsy, hematologic/oncologic diseases, and asthma had the lowest psychosocial health scores and the most significant impact was on the healthy siblings of children with cerebral palsy.

Healthy siblings of children with a chronic illness may experience a loss of appetite, eating disorders, loss of weight or eating more than necessary, and sleep disorders (Van Riper, 2003; Wilkins and Woodgate, 2005). However, healthy siblings reported only a few physical symptoms when they became sick because of the

severity of their sibling's illness (Van Riper, 2003). In this study, the evaluation of physical health scores showed an impact only among the healthy siblings of children with hematologic/oncologic illnesses.

Evaluations made on combined chronic illnesses are mainly based on standard recommendations and determining their impact on family life rather than the specific effects and consequences of these illnesses. In the present study, a comparison between the healthy siblings of all children with a chronic illness and the control group showed that all QoL scores were low; the impact was more obvious in some illnesses, whereas in others, the scores were similar to those of the control group. This indicates that the illnesses should be evaluated individually during QoL measurement. Studies have shown that the course of illness, having a sibling who requires a wheelchair, and the need for intensive care or treatment at home or in the hospital (e.g., for cancer and cystic fibrosis) have more significant negative impacts on the healthy sibling's QoL (Barlow and Ellard, 2006; Read et al., 2011). This may explain the impact in our cancer and cerebral palsy groups. Since celiac disease only requires a change in diet and no hospitalization or interventional therapies, its impact on the healthy sibling's QoL is likely to be much lower. Although previous studies on the healthy siblings of asthmatic children have shown less impact, our study showed that QoL was significantly affected in these children (Barlow and Ellard, 2006). This suggests that geographic differences may be important in QoL measurements; it also supports the hypothesis that the perception of illness may vary between different populations. Sharpe and Rossiter (2002) showed that the psychosocial impact on healthy siblings of children with a chronic illness that has a high mortality risk (e.g., cancer) was similar to the impact among healthy siblings of children with a chronic illness that has a low mortality risk (e.g., diabetes, gastrointestinal diseases, and asthma). Thus, there is no clear consensus about the impact of the severity of the illness or poor prognosis on the healthy siblings' QoL. The time since diagnosis is likely to have an impact on healthy siblings' QoL as well as on the severity and prognosis of the chronic illness (Alderfer et al., 2010). We found that the psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness were not associated with the duration of the chronic illness. Studies have shown that healthy siblings of children with cancer were most affected within the first month after the diagnosis, and the impact was reduced six months after the diagnosis (Houtzager et al., 2004). The time since diagnosis led to a positive impact on the cancer patients' siblings, but it had no impact on the healthy siblings' QoL in illnesses such as epilepsy or diabetes (Jackson et al., 2008). Some studies have shown that the impact was reduced with older age at diagnosis in healthy siblings of children with epilepsy and diabetes (Hames and Appleton, 2009; Wennick et al., 2009). New studies are required to evaluate the duration after diagnosis, since it varies by type of illness. Our evaluations based on the age groups of healthy siblings of children with a chronic illness showed no difference in parents' reports by age group, whereas in healthy siblings' self-reports, the psychosocial health score was higher in the 13–17 and 7–12 age groups than in the 5–7 age group. The marked psychosocial impact in younger children indicates the importance of studies for support programs for children in this age group.

The QoL scores of healthy siblings of children with a chronic illness were higher in the parent reports than in the healthy siblings' self-reports. This suggests that parents of children with a chronic illness may overlook the impact of the chronic illness on the QoL of their healthy children. On the other hand, in the parent reports, the psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness were significantly lower than those of the healthy siblings of healthy children. In the parent reports, the psychosocial health scores were significantly lower among the healthy siblings of children with cerebral palsy, hematologic/oncologic diseases, asthma, diabetes, celiac disease, and epilepsy than among the healthy siblings of healthy children, but there was no difference in the psychosocial health scores between these conditions. In the presence of a chronic illness in children, the psychosocial impact on healthy children was perceived in a similar manner by parents independent of the type and severity of the illness. The status of children or adolescents and their siblings has often been described through parents' reports, which may result in the overlooking of changes in QoL among healthy siblings, as shown by our study. In other words, the world of healthy siblings has been described by adults, but there are substantial discrepancies between the perspectives of parents and their children. In a meta-analysis by Sharpe and Rossiter (2002), the siblings' self-reports were more positive. Therefore, an evaluation of QoL among healthy siblings would provide more accurate information about their experiences and perceptions.

In most studies, the psychosocial assessment of healthy siblings is based on parent reports [20]. In studies about QoL measurement scales, Varni *et al.* (2003 and 2007) stated that a total health score of less than 71.44 in child self-report scales and less than 67.44 in parent proxy-report scales indicated an "impaired QoL." When assessing the impact on children who grow up with a sibling affected by a chronic illness, it is important to measure the relationship between the siblings from the healthy siblings' perspective in addition to QoL evaluations based on the adults' perspective (Knecht et al., 2015). In the healthy children's self-reports, 30.4% of the healthy siblings of children with a chronic illness had an impaired QoL—higher than in the group of healthy siblings of healthy children. In the parent reports, no impairment was observed in the healthy control group, but impairment was observed in 15.1% of the healthy siblings of children with a chronic disease. The level of impaired QoL was

lower in the parent reports than in the healthy children's self-reports, indicating that QoL impairment was less noticed by parents.

The impact on the QoL of healthy siblings was more remarkable in cases with a severe illness and disability. In the children's self-reports, the prevalence of impaired QoL was 42.1%, 51.6%, 45.0%, and 30.7% in the cerebral palsy, hematologic/oncologic disease, asthma, and epilepsy groups, respectively—all higher than in the control group (14.1%). The prevalence of impaired QoL in healthy siblings was lower in the diabetes group (9.9%) and celiac disease group (3.3%) compared to the control group. Although the literature has available data on the healthy siblings of children with cancer, data about the healthy siblings of children with other chronic illnesses are limited. Although there is no difference in bodily functions between the healthy siblings of cancer patients and their peers, they feel anxious about their health status (Alderfer et al., 2010). Murray *et al.* (1999) reported that healthy siblings have a fear of developing the same illness their sibling has.

Our studies have some limitations. These study aim to evaluate the QoL of healthy siblings of children with chronic diseases, however we did not perform disease severity for each disease, especially for children with cerebral palsy due to low number of subgroups. While we enrolled children with epilepsy without nonmotor retardation, seizure frequencies might also affect the parent's and sibling's QoL. Also other disease (especially Celiac disease and diabetes) related factors might affect the QoL, further studies could evaluate these situations. Studies on the evaluation of QoL in healthy siblings of children with a chronic illness are limited. Our study is the first to include more than one group of conditions and assess the QoL from the perspective of both children and parents. Studies on healthy siblings of children with a chronic illness are usually based on an evaluation of the QoL indexes of both the ill child and the healthy sibling; few studies have compared the healthy siblings of children with a chronic illness and the healthy siblings of healthy children. Since different therapeutic approaches, the time since diagnosis, and the severity of illness/mortality may have different impacts on the QoL of healthy siblings of children with a chronic illness, studies on children with a chronic illness should address the chronic illness individually by category (Elissa Lampe Deggelman, 2011). Studies have shown that poor psychosocial functions observed in healthy siblings due to a chronic illness or disability also extended into their adulthood (Barlow and Ellard, 2006; Sharpe and Rossiter, 2002; O'Neill and Murray, 2016; Williams et al., 2009; Stoneman, 2005). The relationship between siblings is influenced by their cognitive, social, and emotional development, which are determinants of QoL throughout their lives (Verte et al., 2003). Considering the frequency of chronic illnesses and increased life expectancy, it is necessary to evaluate the impact of a child's chronic illness on the child's healthy siblings at home, mainly the psychosocial impact on their global QoL. The inclusion of healthy siblings in the support programs for parents of children with a chronic illness may be beneficial (Ahola Kohut et al., 2017; Jackson et al., 2016), since brotherhood/sisterhood is a lifelong relationship that determines one's identity and personality during adulthood.

KEY MESSAGES

- The diagnosis of a physical chronic condition during childhood is a source of serious stress in the family and also has psychosocial impacts on the siblings of the children with a chronic illness.
- The physical health, psychosocial health, and total health scores of healthy siblings of children with a chronic illness were significantly lower than those of siblings of healthy children.
- In healthy siblings, we observed a global impact on the HrQoL, including psychosocial scores, and a low level of parental awareness about this situation. This might increase the risk of emotional neglect and abuse in these children.

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Table 1. Demographical findings of children with chronic disease and healthy siblings of children with chronic disease

	N	Age (months) Mean ± SD	Gender B/G	Disease duration (months)	Age of Healthy Siblings (ay)	Gender of healthy siblings B/G	Percentage of older healthy siblings than children with CD
Chronic disease TOTAL	191	116 ± 56	85/106	36	119 ± 58	102/89	101 (%52.9)
Cerebral Palsy	26	108 ± 60	17/9	72	119 ± 60	17/9	16 (%61.5)
Hematologic Oncologic Disorders	39	105 ± 48	16/23	24	124 ± 57	19/20	14 (%35.9)
Asthma	31	98 ± 51	17/14	24	98 ± 59	14/17	14 (%45.2)
Type 1 Diabetes	28	145 ± 46	7/21	36	125 ± 59	18/10	12 (%42.9)
Celiac disease	33	143 ± 48	11/22	48	133 ± 53	20/13	15 (%45.5)
Epilepsy	34	104 ± 65	17/17	24	115 ± 60	14/20	19 (%55.9)

Table 2. The psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness and the healthy siblings of healthy children (Self-reports of healthy siblings)

	Healthy siblings of children with a chronic illness (n=191)	Healthy siblings of healthy children (n=100)	p
Psychosocial health score	72.6 ± 13.7 (25-100)	82.1 ± 11.6 (56-100)	<i>p</i> < .001
Physical health score	82.6 ± 13.8 (38-100)	87.2 ± 8.8 (63-100)	<i>p</i> < .01
Total health scores	75.1 ± 11.6 (40-97)	83.4 ± 10.4 (60-100)	<i>p</i> < .001

*Health scores were expressed as mean ± SD (range; minimum and maximum)

Table 3. The psychosocial health, physical health, and total health scores of the healthy siblings of children in the chronic illness subgroups and the healthy siblings of healthy children (Self-reports of healthy siblings)

	Cerebral Palsy (n=26)	Hematologic Oncologic Disease (n=39)	Asthma (n=31)	Type 1 Diabetes (n=28)	Celiac Disease (n=33)	Epilepsia (n=34)	Healthy Controls (n=100)
Psychosocial health score	69.7 ± 15.8 (25-96)	66.2 ± 11.1 (33-81)	69.9 ± 16.3 (31-90)	75.9 ± 8.0 (56-88)	82.1 ± 7.4 (58-91)	73.4 ± 15.7 (38-100)	82.1 ± 11.6 (56-100)
Physical health score	86.8 ± 13.2 (63-100)	79.4 ± 9.4 (48-83)	76.9 ± 19.7 (43-100)	83.2 ± 11.5 (46-96)	84.3 ± 6.8 (71-96)	86.0 ± 16.4 (46-100)	87.2 ± 8.8 (63-100)
Total health scores	74.0 ± 13.6 (43-98)	69.5 ± 9.4 (48-83)	70.8 ± 14.6 (40-93)	77.7 ± 6.6 (62-87)	82.7 ± 6.6 (67-91)	76.5 ± 13.3 (40-97)	83.4 ± 10.4 (60-100)

Table 4. The psychosocial health, physical health, and total health scores of the healthy siblings of children with a chronic illness and the healthy siblings of healthy children (Parent Reports for Healthy Siblings)

	Healthy siblings of children with a chronic illness (n=191)	Healthy siblings of healthy children (n=100)	p
Psychosocial health score	76.8 ± 12.3 (40-100)	88.1 ± 6.6 (71-100)	<i>p</i> < .001
Physical health score	80.7 ± 15.4 (28-100)	87.6 ± 8.5 (65-100)	<i>p</i> < .001
Total health scores	77.7 ± 11.3 (44-97)	88.0 ± 6.0 (74-100)	<i>p</i> < .001

*Health scores were expressed as mean ± SD (range; minimum and maximum)

Table 5. The psychosocial health, physical health, and total health scores of the healthy siblings of children in the chronic illness subgroups and the healthy siblings of healthy children (Parent Reports for Healthy Siblings)

	Cerebral Palsy (n=26)	Hematologic Oncologic Disease (n=39)	Asthma (n=31)	Type 1 Diabetes (n=28)	Celiac Disease (n=33)	Epilepsia (n=34)	Healthy Controls (n=100)
Psychosocial health score	77.0 ± 15.0 (45-100)	76.1 ± 12.2 (40-96)	77.9 ± 14.4 (44-92)	75.3 ± 12 (53-100)	81.5 ± 7.1 (68-95)	75.1 ± 13.0 (48-96)	88.1 ± 6.6 (71-100)
Physical health score	83.3 ± 17.8 (28-100)	78.2 ± 16.2 (37-100)	81.5 ± 15.3 (53-100)	76.6 ± 16.1 (28-100)	83.4 ± 10.2 (46-96)	81.1 ± 15.2 (31-100)	87.6 ± 8.5 (65-100)
Total health scores	78.6 ± 13.7 (40-100)	76.9 ± 11.0 (44-95)	79.3 ± 11.6 (58-92)	75.0 ± 13 (38-100)	82.0 ± 6.9 (62-95)	76.6 ± 11.3 (44-97)	88.0 ± 6.0 (74-100)

Table 6. Prevalence of Impaired QoL in Healthy Siblings of Children with a Chronic Illness and Healthy siblings of healthy children

	Healthy siblings of children with a chronic illness (n=191)	Healthy siblings of healthy children (n=100)	p
Child's total self-reported score of less than 71.44	45/148 30.4%*	11/78 14.1%	<i>p</i> = .005
Parent-report score of less than 67.44	29/191 15.1%	0/100	<i>p</i> < .001

*The parents reported less impairment than the healthy siblings did (30.4% vs. 15.1%, respectively, *p*<0.05),

Table 7. Prevalence of Impaired QoL in the healthy siblings of children in the chronic illness subgroups and healthy siblings of healthy children

	Cerebral Palsy (n=26)	Hematologic Oncologic Disease (n=39)	Asthma (n=31)	Type 1 Diabetes (n=28)	Celiac Disease (n=33)	Epilepsia (n=34)	Healthy Controls (n=100)
Child's total self-reported score of less than 71.44	8/19 42.1%	16/31 51.6%	9/20 45%	3/22 9.9%	1/30 3.3%	8/26 30.7%	11/78 14.1%
Parent-report score of less than 67.44	7/26 26.9%	4/39 10.2%	8/31 25.8%	4/28 14.2%	1/33 3.0%	5/34 14.7%	0/100 0

Uncorrected Proof