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Belgian siblings of children with a chronic illness: Is their quality of life different from their peers?

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Abstract

To assess Belgian siblings' self-reported quality of life (QoL) and the impact of illness on four different paediatric illnesses. Healthy siblings ($n = 131$) of children with type 1 diabetes, cancer, congenital heart disease (CHD) and cystic fibrosis (CF) completed the Child Health Questionnaire and the Sibling Perception Questionnaire. Results were compared to those of a matched group of siblings of healthy children. Siblings reported a good QoL, similar to controls, with the exception that siblings reported better on the QoL domain pain ($p < .01$). QoL was not related to time since diagnosis but the impact of illness was higher nearer to the time of diagnosis ($r = -.39$, $p < .001$). QoL of siblings of children with CHD or cancer was lower than QoL in the CF or type 1 diabetes group whilst impact of illness was highest for the CHD group. QoL of siblings of a child with a chronic illness is similar to the QoL of peers. Studies investigating siblings' QoL or the impact of illness on siblings should include the day-to-day demands of the illness as well as less obvious illness-related issues like 'hidden stress' and 'sense of control'.

Keywords

Chronic illness, impact of illness, quality of life, self-report, siblings

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Introduction

Progress in paediatric medicine has resulted in an increase in the number of children living with chronic illness, leading to a growing awareness of the changing impact of chronic illness on the family and specifically on siblings (Halfon and Newacheck, 2010). Most studies on such siblings have focused on their adaptation and adjustment to the illness and on the incidence of psychosocial problems (Alderfer et al., 2010; Barlow and Ellard, 2006; Fanos et al., 2005; Gerhardt et al., 2012; Houtzager, 2004; Lobato and Kao, 2002; Nolbris et al., 2007; Sharpe and Rossiter, 2002; Tsuchie et al., 2006; Williams et al., 2002). A recent meta-analysis (Vermaes et al., 2011) found a small negative effect of chronic health conditions (CHCs) on siblings: more internalizing and externalizing problems and fewer positive self-attributions. On the other hand, several studies have described effects on siblings that were labelled positive by the investigators; for example, they may be more aware of illness and disability and more caring towards others and they may take on more responsibility (Alderfer et al., 2010; Barlow and Ellard, 2006; Bellin and Kovacks, 2006; Harder and Bowditch, 1982; Havermans and Eiser, 1994).

Over the past years, the introduction of the concept of family quality of life (QoL) has broadened sibling research (Alderfer et al., 2010; Demerval et al., 2009; Gundlach et al., 2006; Houtzager, 2004; Houtzager et al., 2001). QoL is a broad-ranging concept affected in a complex way by a person's physical health, psychological state, level of independence, social relationships, personal beliefs and their relationship to salient features of their environment (World Health Organisation (WHO), 1994). Siblings are able to self-report on their QoL, and it was shown to be an appropriate concept to describe siblings' experiences (Moyson and Roeyers, 2012).

The aim of the present study was to assess self-reported QoL and the perception of the impact of a chronic illness on siblings. We have tackled some of the issues raised by Vermaes et al. (2011) concerning the methodology of studies by including a matched control group, a narrow range in the age of the siblings and siblings' self-report. In addition, the study adopted both a categorical and a non-categorical perspective.

The emphasis of the non-categorical approach, described by Stein and Jessop (1982), is that similarities between illnesses outweigh the differences. The rationale is that, in studying life experiences of children, different illnesses should be included in a sample rather than focusing on a single disease. The non-categorical approach is particularly useful when only smaller numbers of patients are available and when there is a need to identify overall themes to improve and develop care programs.

The categorical approach focuses on context-specific characteristics experienced by children and families. Central to this are the specific diagnosis and treatment elements, such as whether the illness requires daily treatment or whether the illness is predictable. This approach is useful for identifying, developing and delivering specific services to children and their families, taking into account disease and treatment (Ingersk et al., 2010). The value of a categorical approach in sibling research has been supported by findings that different illnesses can have a different impact on siblings (Bluebond-Langner, 1996; Houtzager et al., 2001; Sharpe and Rossiter, 2002; Vermaes et al., 2011).

Using the non-categorical approach, we established four hypotheses.

Hypothesis 1: Siblings rate their QoL lower than controls (Sharpe and Rossiter, 2002; Vermaes et al., 2011);

Hypothesis 2: Younger siblings report a better QoL compared to older siblings (Ferrari, 1984; Hollidge, 2001; Houtzager et al., 1999; Houtzager et al., 2003; Sargent et al., 1995; Vermaes et al., 2011);

Table 1. Demographic details of siblings.

Condition	Age mean (SD)	Gender		Time since diagnosis; M (SD), range in years
		Boys	Girls	
CHD	14.3 (2.2)	8	13	13.4 (4.0)*, 6–18
Diabetes	13.2 (2.2)	22	13	4.8 (3.2), 1–14
CF	13.2 (2.1)	15	23	11.9 (2.8)*, 5–18
Cancer	13.3 (2.5)	23	14	3.0 (1.9), 1–8
Total illness groups	13.4 (2.2)	68	63	7.7 (5.2), 1–18

CHD: congenital heart disease; CF: cystic fibrosis.
*The CHD and CF groups were diagnosed a significantly longer time ago than the cancer and diabetes groups ($p < .001$).

Hypothesis 3: Female siblings report a lower QoL than males do (Houtzager et al., 2003, 2004) and

Hypothesis 4: Longer time since diagnosis is related to higher QoL and lower impact of illness.

Hypothesis 4 is based on the finding that time since diagnosis is a moderator of sibling adjustment. Previous studies have shown that siblings respond differently to illness depending on the time since diagnosis (Alderfer et al., 2010; Houtzager et al., 2004).

Using the categorical approach, differences in self-reported QoL and impact of illness between the illness groups were examined. Different illnesses and prescribed treatments produce different day-to-day demands (Sharpe and Rossiter, 2002). From clinical practice, we know that children with diabetes and cystic fibrosis (CF) need intense daily treatment and monitoring. For these children, their days are organized around treatment modalities they cannot (or should not) miss, including physiotherapy for CF patients and daily blood sugar monitoring and insulin injections for patients with diabetes. We established a fifth hypothesis that due to specific day-to-day illness demands, siblings of children with diabetes and CF will report lower on domains of QoL and higher on the impact of the illness than siblings of children with cancer or congenital heart disease (CHD).

Methods

Participants

The participants were siblings of children with four different chronic conditions: cancer, type 1 diabetes (referred to from here on as 'diabetes'), CHD and CF. For brevity, the terms 'sibling(s)' and 'controls' will be used, respectively, for sibling(s) of a chronically ill child and the matched control sibling(s) of a healthy child. The sample is a convenience sample as the four chosen illnesses were those available to the researchers.

Table 1 shows the number of siblings in the different illness groups. The group with diabetes consisted of children with a mean glycosylated haemoglobin a1C (Hba1C) of 7.50 (SD = .9, range 6.3–9.7; Hba1C serves as a marker for average blood glucose levels over the previous 3 months). These children were treated with daily insulin and had to follow a strict dietary regime. The group with CF consisted of children undergoing daily physiotherapy, enzyme replacement therapy, high-calorie dietary intake, daily nebulizing therapy and regular antibiotics treatment. The group with

CHD consisted of children born with serious heart defects, namely tetralogy of Fallot ($n = 11$) and univentricular heart ($n = 10$). All these children had undergone at least one major heart operation. Some children ($n = 10$) took daily medication. The group with an oncological problem ($n = 37$) consisted of 6 children with brain tumour/astrocytoma, 20 children with acute lymphoblastic leukaemia/lymphoblast and 11 with solid tumours. The majority (32) of these children were in follow-up (outpatient clinics), whilst the remaining 5 were on maintenance treatment with medication. The children in follow-up were between 0 and 5 years post active treatment; 20 had physical or neurological/cognitive problems to varying degrees caused by the cancer and/or treatment.

Procedure

Siblings had to be between the ages of 10 and 18 years because the QoL questionnaire used in this study is designed for children of this age range. Siblings also had to be native Flemish speaking. Excluded were families known by the researchers to include someone suffering from psychosocial or psychiatric problems (e.g. maternal depression), because these psychosocial situations would be expected to confound the self-report of the siblings. Siblings reported on their own health, and none of the siblings had to be excluded because of a CHC. No other data were obtained concerning health or behavioural problems.

During regular outpatient clinics, parents were asked permission for their child to participate in the study. For families with more than one sibling, only the sibling closest in age to the Ill child was asked to participate, to avoid overburdening the family with the questionnaires. The consenting parents took the questionnaires home for the sibling to complete; the siblings were asked to read and sign an assent form, and the questionnaires were returned in pre-addressed stamped envelopes.

For the CF group, 100% of the families who consented returned the questionnaire. Several parents ($n = 17$) needed a reminder by phone. Response rates for the other groups were 86% for the CHD group, 87% for the oncology group and 92% for the diabetes group. Reasons for drop out were not collected and it was not possible to compare responders with non-responders.

Control group

Wuytack (2008) collected data from 437 children who completed the Child Health Questionnaire – Child Form (CHQ-CF87) during a school day under the supervision of their teacher. Prior to this, informed consent was obtained from parents and assent from the children. Data on health in the family were self-reported and children who had a chronic condition or a sibling with a chronic illness (mostly asthma or diabetes) were excluded from the matching. For the present study, 131 siblings were perfectly matched according to age and sex with controls from the Wuytack's study (Table 1).

Ethics

The study was approved by the hospital ethics committee. The committee specifically looked at the questions that were asked, the way data were processed with regard to confidentiality and the informed consent and assent forms. The committee follows the guidelines for Good Clinical Practice and the Declaration of Helsinki to protect individuals participating in studies (http://www.wma.net/en/30publications/10policies/b3/).

Table 2. The CHQ-CF87 subscales: description of domains (of lower score).

CHQ-CF87 subscale (no. of items)	Interpretation of lower score	Cronbach's α; $n = 262$
Physical functioning (9)	Child is restricted in fulfilling physical activities including self-care due to health	.71
Role/social restrictions – emotional (3)	Child is restricted in school or social activities due to emotional problems	.84
Role/social restrictions – behaviour (3)	Child is restricted in school or social activities due to behavioural problems	.73
Role/social restrictions – physical (3)	Child is restricted in school or social activities due to physical problems	.78
Bodily pain (2)	Child has extreme, serious, frequent and debilitating pain	.93
Behaviour (17)	Child behaves aggressive, immature and delinquent	.84
Mental health (16)	Child is anxious and depressed	.87
Self-esteem (14)	Child is dissatisfied with his/her abilities, appearance, family relations and social relations and life in general	.91
General health (12)	Child believes that his/her health is bad and will deteriorate	.72
Family activities (6)	The health of the child restricts or disrupts family activities or is a source of family tension	.82
Family cohesion (1)	Family relations are rated as very bad	

CHQ-CF87: Child Health Questionnaire – Child Form.

Measures

Demographic and illness variables. Age and sex were obtained via the questionnaire. Time since diagnosis was identified from medical records.

Questionnaires. Quality of Life: CHQ-CF87. The CHQ-CF87 (Landgraf et al., 1999) is a general questionnaire (86 items) on QoL, assessing physical, social and emotional welfare over the previous 4 weeks of children between the ages of 10 and 18 years, consisting of 10 multi-item subscales and 1 single-item subscale (see Table 2 for a description of the scales). The average scores for the CHQ-CF87 were calculated from 4-, 5- or 6-point scales on each item and transformed into a 0–100 scale, where a higher score indicates better QoL. The CHQ has previously been used in Flemish samples (Joos et al., 2001; Wuytack, 2008), making it a good instrument to use in the present study. Good reliability has been reported (Joos et al., 2001; Landgraf et al., 1999; Norrby et al., 2003; Wuytack, 2008). Reliability of the current sample is shown in Table 2.

Impact of illness: Sibling Perception Questionnaire (SPQ). The original version of the SPQ was developed and described by Carpenter and Sahler (1991) to assess the impact of cancer on siblings. Lobato and Kao (2002) adapted this questionnaire for use with siblings of children with other diseases. Their version was translated into Flemish by a translation agency using both forward translation and back translation, changing the word 'problem' into 'illness'. The adapted SPQ consists of 22 items, on a scale of 1 (*never*) to 4 (*often*). Following previous studies (Guite et al., 2004; Lobato and Kao, 2002; Sloper and While, 1996), scores for these items were summed to create a Negative Impact Composite Scale. Examples of such items include 'I feel sad about the illness of my brother/sister' and 'I wish my parents would spend less time with my brother/sister'. The Negative Impact Composite Scale was used in subsequent analyses, with higher scores

Table 3. Mean domain scores of the CHQ-CF87 scale of siblings and of controls.

CHQ-CF87 domain	Siblings; M (SD)	Control; M (SD)	Cohen's *d*	*t*
Physical functioning	97.00 (5.99)	95.96 (7.02)	−.16	−1.29
Role/social restrictions – emotional	92.96 (12.03)	91.43 (17.78)	−.10	−.81
Role/social restrictions – behaviour	95.17 (10.41)	94.57 (12.35)	−.05	−.42
Role/social restrictions – physical	98.30 (6.81)	96.27 (1.36)	−.23	−1.87
Bodily pain	80.38 (18.57)	74.89 (16.47)	.31	−2.53*
Behaviour	82.32 (10.70)	81.91 (11.13)	−.03	−.30
Mental health	76.66 (12.43))	76.80 (14.05)	.00	.08
Self-esteem	77.52 (11.27)	75.80 (13.43)	−.13	−1.12
General health	76.66 (14.00)	73.55 (14.89)	−.21	−1.73
Family activities	86.99 (15.03)	84.57 (17.05)	−.15	−1.21
Family cohesion	73.28 (21.04)	77.29 (22.46)	.18	1.49

CHQ-CF87: Child Health Questionnaire – Child Form.
*$p < .01$

reflecting a greater impact of illness (range 1–4). The Cronbach's α value of the Negative Impact Composite Scale for the current sample was .82.

Statistical analyses

Data analyses were conducted using SPSS 19. Descriptive statistics were used to examine the demographic characteristics of the samples. The non-parametric Kruskal–Wallis test was used to test the differences between the groups (matching procedure). Pearson's correlations were used to assess correlations between age and time since diagnosis. To investigate group differences, Student's *t* tests ($p < .05$) and analyses of variance were applied to investigate group differences and sex and age differences between the groups. Cohen's *d* was calculated to measure the strength of the relationship between the variables. For hypothesis 3, analyses of variance were used with post hoc analysis (Tukey's test) and $p < .05$ to investigate pair wise comparison.

Results

Non-categorical approach

Siblings rated their QoL generally higher than controls, although this was statistically significant only for the domain bodily pain (Table 3), with siblings reporting fewer aches and pains than controls. Cohen's *d* was low, indicating a weak effect. No other significant differences between groups were found in the scores for the other domains.

Age and sex

No differences between the main groups (siblings vs. control) were found in the CHQ domain for age or sex. For the combined illness groups, girls scored lower than boys in the CHQ mental health domain (79.43 vs. 73.81, $p < .01$) and age showed an inverse relationship with self-esteem ($r = -.34$, $p < .001$), indicating that older siblings reported lower on the domain self-esteem than younger siblings.

Table 4. Partial Pearson's correlations between time since diagnosis and scores on the Negative Impact Composite Scale and scores on the quality of life domains.

	Time since diagnosis	Time since diagnosis (controlling for illness)
Negative Impact Composite Scale	−.39**	−.29**
Physical functioning	.24	.04
Role/social restrictions – emotional	.01	.01
Role/social restrictions – behaviour	.05	.01
Role/social restrictions – physical	.17	.11
Bodily pain	.02	.11
Behaviour	.05	.01
Mental health	.04	.09
Self-esteem	−.09	−.08
General health	.06	.18
Family activities	.15	.12
Family cohesion	−.06	−.01

**$p < .001$.

Time since diagnosis

The mean time since diagnosis was 7.7 years (SD = 5.2, range 1–18 years). As expected, higher negative impact scores were associated with a shorter time since diagnosis ($r = -.42$, $p = .001$). QoL scores were not related to time since diagnosis (Table 4). Because of the significant difference between the illnesses in time since diagnosis (Table 1), a categorical approach was adopted to investigate further such differences. Partial correlations, controlling for illness, showed that a shorter time since diagnosis was associated with higher scores on the Negative Impact Composite Scale ($r = -.29$, $p < .01$) but not with scores in domains of QoL (Table 4).

Categorical approach

Table 5 presents mean scores for QoL and negative impact for each of the four illness groups. The analyses of variance showed a significant difference in mean scores in the domains behaviour, mental health, self-esteem and the Negative Impact Composite Scale. Post hoc tests were performed to investigate all pair wise comparisons between group means for these scales. Contrary to hypothesis 5, siblings of children with CF scored higher than siblings of children with cancer ($p < .05$) and CHD ($p < .05$) in the domain behaviour. Additionally, for the domain mental health, siblings of children with CF ($p < .01$) and diabetes ($p < .05$) scored higher than siblings of children with CHD. For self-esteem, siblings of children with diabetes scored higher than siblings of children with CHD ($p < .05$). For the Negative Impact Composite Scale, post hoc analyses showed that siblings of the children with cancer reported a higher impact than siblings of children with CHD ($p < .001$), CF ($p < .01$) and diabetes ($p < .05$). Other comparisons were not significant.

Discussion

Based on a non-categorical approach, siblings of children with four different illnesses were joined and matched with a control group. Contrary to expectations, siblings reported a good QoL, similar

Table 5. Analysis of variance comparing mean domain scores of CHQ-CF87 between the four illness groups (CHD = 21, diabetes = 35, CF = 38 and cancer = 37).

	Condition	M	SD	*F*
Physical functioning	CHD	97.35	3.13	.99
	Diabetes	95.66	6.75	
	CF	98.05	5.69	
	Cancer	96.99	6.69	
Role/social restrictions – emotional	CHD	91.53	14.86	.25
	Diabetes	93.65	10.53	
	CF	93.85	11.46	
	Cancer	92.19	12.52	
Role/social restrictions – behaviour	CHD	97.88	5.68	1.50
	Diabetes	96.50	9.24	
	CF	95.02	9.56	
	Cancer	92.49	13.62	
Role/social restrictions – physical	CHD	100.00	.00	1.32
	Diabetes	98.09	8.29	
	CF	99.12	3.03	
	Cancer	96.69	9.39	
Bodily pain	CHD	77.14	17.36	.64
	Diabetes	79.14	15.97	
	CF	80.78	17.76	
	Cancer	82.97	15.25	
Behaviour	CHD	78.22	10.10	4.03**
	Diabetes	84.83	8.86	
	CF	85.36	8.75	
	Cancer	79.14	12.97	
Mental health	CHD	68.97	13.40	4.74**
	Diabetes	79.46	10.50	
	CF	79.93	11.03	
	Cancer	75.00	13.14	
Self-esteem	CHD	71.59	12.85	3.25*
	Diabetes	80.61	10.41	
	CF	78.90	10.43	
	Cancer	76.54	10.95	
General health	CHD	73.84	18.03	.33
	Diabetes	77.51	13.82	
	CF	77.10	11.95	
	Cancer	76.98	13.94	
Family activities	CHD	87.89	14.18	1.94
	Diabetes	85.00	16.5	
	CF	91.44	10.11	
	Cancer	83.78	17.42	
Family cohesion	CHD	66.19	24.38	1.07
	Diabetes	73.71	18.91	
	CF	76.31	20.52	
	Cancer	73.78	21.42	
Negative Impact Composite Scale	CHD	1.82	.38	8.61***
	Diabetes	2.09	.36	
	CF	2.00	.33	
	Cancer	2.33	.45	

CHD: congenital heart disease; CF: cystic fibrosis; CHQ-CF87: Child Health Questionnaire – Child Form.
***$p < .001$; **$p < .01$; *$p < .05$.

to controls, and significantly better scores in the domain bodily pain, indicating that siblings report fewer aches and pains. These results may be used in clinic to reassure parents who are worried about the effect of an illness on siblings. The bodily pain effect is not strong and the result should be interpreted with caution. One could hypothesize that the concept of bodily pain has a different meaning for siblings of chronically ill children. Herrman (2010) described reactions to pain in siblings of children with diabetes, with some expressing sympathy for their sibling for undergoing painful procedures, while others stated that the pain was not so bad. It may be that the siblings of sick children perceive their bodily sensations within a different framework of reference for pain in comparison with siblings of healthy children who are less likely to have been confronted with illness-related pain. Other researchers have hypothesized an alternative explanation that siblings report less physical pain or problems so as not to cause distress to their parents (Hollidge, 2001; Sharpe and Rossiter, 2002).

The meta-analyses by Sharpe and Rossiter (2002) and Vermaes et al. (2011) showed that the sibling literature is inconclusive with regard to age and sex differences. Studies are difficult to compare or draw conclusions from as they use different instruments to assess well-being or adjustment. For the present study, only two findings confirmed the hypotheses: the older siblings report lower on the domain self-esteem than the younger siblings, and sisters report lower on the domain mental health compared to brothers. Both these results were described in studies on cancer and CF (Houtzager et al., 2003, 2004, Wennström et al., 2005), concluding that older siblings, especially girls, were at risk of developing lower QoL.

Previous research has shown the importance of time since diagnosis (Alderfer et al., 2010; Houtzager et al., 2004; Williams, 1997). Alderfer et al. (2010) reported that sibling outcomes are more likely to differ from controls nearer to the time of diagnosis. It is possible that siblings become used to the illness and treatment over time and gain a level of resilience against the stress (Williams, 1997); life may normalize over time. This may explain why no association was found between the domains of QoL and time since diagnosis, because the sample only included siblings whose sister or brother had been diagnosed at least one year ago. However, the results did show that the perceived impact of the illness was higher nearer to the time of diagnosis. These findings suggest that self-reported QoL is more stable, whilst the perceived impact of the illness varies over time. These findings promote the use of both general QoL and illness-related assessment to cover both general and illness-related aspects of concern to siblings (Alderfer et al., 2010; Hartling et al., 2010). Illness may have a great impact on the lives of siblings, especially shortly after diagnosis, but not necessarily a huge impact on their QoL. When counselling families, this might be taken into account in interpreting siblings' behavioural changes over time.

For the final hypothesis, the comparative approach was used to evaluate the differences between the four illness groups. The hypothesis was that siblings of children with diabetes and CF would report lower in domains of QoL than siblings of children with CHD or cancer. This hypothesis was based on the assumption that the lives of siblings of children with diabetes and CF were more disrupted by the illness due to the prescribed treatment regimen on a day-by-day basis (including medicine intake, physiotherapy, nebulizing, injections, diet, etc.). However, the results did not support this hypothesis. Siblings of children with CHD or cancer reported more behavioural and internalizing problems than siblings of children with CF and diabetes. From the descriptions in the CHQ, these siblings worry more, are less satisfied with their abilities and may be more immature. An explanation may be that the hypothesis does not take into account the role of (masked) stress, worry and life threat, and it disregards the impact of information, beliefs and communication about an illness. Cancer is often more negatively perceived in our society than, say, diabetes as it is

closely related to death. These themes contribute to siblings' representations of illness and coping (Alderfer et al., 2010; Houtzager, 2005). Siblings of children with diabetes and CF are confronted with the illness and treatment on a daily basis, which may actually provide some sense of control. These siblings see and understand what is going on, and they have learned what they can do to help in the care of their brother or sister (Herrman, 2010).

Although children with cancer or CHD, like those with CF and diabetes, are in regular follow-up, in the 'day-to-day care' of children with cancer, there is relatively little control that prevents the illness from reoccurring. Moreover, there are often no direct, obvious signs of relapse. Children with CHD cannot be sure that their cardiac condition will remain stable as they grow older, possibly even needing new surgery. Siblings of children with CHD or cancer may report more internalizing problems because they worry more about the risk of recurrence or unexpected problems. The present results support the view that in the development of support programs for siblings, it is important to consider carefully *which* specific interventions may help *which* group of siblings most (Hartling et al., 2010). A good example of such a specified program is the bead program for siblings of children with chronic childhood heart disease (Redshaw and Wilson, 2012). Furthermore, the present data support the conclusion by Malcolm et al. (2013) that sibling support should be cognisant of the trajectory of the illness.

Research often shows inconsistent results due to methodological differences and diverse approaches (Alderfer et al., 2010; Houtzager et al., 2004; Vermaes et al., 2011). In the present study, we tried to overcome some of the methodological difficulties mentioned in the introduction by clarifying the approach that was used and by including siblings of children with multiple illnesses and a control group perfectly matched for age and sex. We included a group of children in the preadolescent to adolescent age range in order to obtain a homogeneous developmental group.

The findings present interesting challenges to our clinical care practice. For example, it is important to know that the impact of an illness is highest nearer to the time of diagnosis, but QoL seems relatively stable over time. In addition, siblings of children with an illness that requires intensive day-to-day care and treatment do not report a worse QoL compared to siblings of children with an illness that demands less active treatment.

Nevertheless, a number of limitations remain. First of all, the sample comes from only one hospital in one country. Future studies may need to focus on how the cultural background of siblings may influence the impact of a chronic illness on their siblings. Second, most of the recruited families participated but no information was collected on the non-responders. The siblings completed the questionnaires at home, for convenience. This made it impossible to ascertain the extent to which siblings were influenced by parents or others when filling in the questionnaire, or whether parents checked the forms, with implications for confidentiality and reliability of self-reports. Although the questionnaire was carefully chosen to assess QoL in siblings, a number of domains showed a ceiling effect that made the results of these domains less valuable. The four illness groups were relatively small and, especially those of children with CHD and cancer, were heterogeneous as subsamples, which limit the generalizability of the data. The power of the interactions was limited and future research should aim to include larger and more homogeneous samples. Furthermore, the effects of other individual or contextual factors that may be related to self-reported QoL, birth order, family functioning, quality of sibling relationships, and so forth were not explored and controlled for.

In future studies, it would be important to understand more about the impact of the day-to-day burden of an illness in comparison with the possible burden of underlying emotions and worries. Longitudinal research is important for greater understanding of the relationship between time since diagnosis and QoL and the impact of illness.

Clinical implications

1. Siblings of children with a chronic illness report a better QoL compared to their peers, indicating that chronic illness of a child does not automatically predispose siblings to report a lower QoL. Parents are often worried about the effect of an illness on siblings' well-being. Health-care professionals may cautiously use the present data to help parents overcome their worries.
2. Siblings of children who have recently been diagnosed experience a higher impact due to the illness. In counselling families, this might be taken into account when explaining changes in siblings' behaviour over time.
3. One may expect older adolescent siblings to experience more difficulties.
4. The present results suggest that siblings of families with a daily medical routine (diabetes and CF) report a better QoL. Families with a child with chronic illness may be encouraged to use their routine to improve family well-being.
5. Society's concepts of illness, hidden stressors, uncertainty and perceived uncontrollability may affect siblings' lives. When developing support programs for siblings, it is important to take this into account and to consider that different groups may need specific interventions.

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