Consequences of care: Parents of children with a chronic disease
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General Discussion
Several changes in pediatrics and in society have altered the care giving demands for parents of chronically ill children. The prevalence of chronic childhood disease has increased, and the medical advancements enabling complex care to be given in the home-situation, has changed the caring tasks at home. Also, the emphasis on informal care has increased and family demographics have changed. All this has stressed the need for thorough investigation of the consequences of the care for children with a chronic disease for their parents.

This thesis reports the results of the Care-project, which investigated quality of life, employment and leisure time of parents of chronically ill children.

The aim of our study was to explore:

1. **Health Related Quality of Life**,  
2. **employment and leisure activity time**,  
3. to estimate a monetary valuation of wellbeing of parents caring for a chronically ill child in comparison with parents of children who are able to grow and develop in good health, and  
4. to estimate predictors of employment, leisure activity time, and HRQoL.

The conceptual model used to answer these questions described the dynamics of care giving by using HRQoL as an outcome measure, which was explained by socio demographic and disease related variables. In addition to several main effects, a mediating effect of social variables as income, employment, leisure time and emotional support was expected. The mediating variables employment and leisure activity time were also analyzed as dependent variables. In this final chapter a summary (§1) and discussion of the results (§2) is given including discussion of the strengths and limitations of the present study. Furthermore, clinical implications are discussed (§3), as well as recommendations for future research (§4) and a final conclusion is given (§5).

**1 MAIN FINDINGS**

The Care-project was designed to provide answers to the following research questions:

1. **How is the HRQoL of parents of chronically ill children compared to parents of healthy schoolchildren?**  
2. **Which characteristics (socio-demographic, disease related, and social variables) are associated with the HRQoL of parents of chronically ill children?**  
3. **How does chronic illness of a child relate to parental well-being in comparison with parents of healthy children and what are the costs of differences in well-being expressed in monetary value?**  
4. **What is the relationship of being a parent of a child with a chronic disease with parental employment and leisure activity time and which factors (disease related and social) are associated with these outcomes in parents of children with a chronic disease?**
In chapter 2 we described the lower HRQoL scores of parents of chronically ill children compared to parents of healthy children on the following domains: gross motor functioning, cognitive functioning, sleep, pain, social functioning, daily activities, sexuality, vitality, positive emotions, and depressive emotions. Comparison of disease specific groups with the comparison group mainly showed lower HRQoL regarding social and daily functioning, vitality and sleep, and having less positive and more negative emotions. Especially parents of children with metabolic diseases and sickle cell disease in had lower scores on a large number of domains. These outcomes were studied more thoroughly. Almost half of all parents were at risk for HRQoL impairment. In chapter 6 we focussed on the mothers of children with sickle cell disease, a group with low HRQoL scores and a lower socio-economic status than parents in the other groups in the Care-project (including the comparison group). The HRQoL scores of these mothers showed fewer differences when compared to mothers from a more similar socio-economic background. However, mothers of children with sickle cell disease still report a lower HRQoL regarding depressive moods, daily activities and vitality.

In our model, presented in chapter 3, parental HRQoL was directly negatively associated with female gender, lower parental age, lower educational level, higher care-dependency of the child, having a chronic illness as a parent, less emotional support, and number of days parents went on holiday in the past year. In addition to these direct effects, the effect of educational level, having a partner, being born in the Netherlands, being chronically ill, having a chronically ill partner and care-dependency on HRQoL was mediated by holiday and emotional support.

Within the group of parents of children with metabolic disorders (chapter 7), psychosocial determinants (e.g. loss of friendship and emotional support) appeared to be more important determinants of parental HRQOL than socio-demographic and disease related variables.

In chapter 4, employment and leisure activity time of parents were addressed. Parents of chronically ill children worked fewer hours a week, mothers less often participated in the labor force, and parents spent less time doing leisure activities than parents of healthy children. Care dependency of the child was negatively associated with family employment and maternal labor force participation, but not with leisure activity time. Parents using child care facilities had a higher probability of working more hours a week. In addition, the effect of child care on employment was stronger for mothers than on a family level. A low educational level was a risk factor for family and maternal employment and for leisure activity time.

In chapter 5 we showed that having a chronically ill child had a negative impact on parental subjective well-being. This impact was larger for parents having a child with a progressive illness followed by parents whose child had a relapsing illness.

For the specific disease groups, the effect on well-being was highest for parents of children with Duchenne and multiple complex handicaps. The monetary value equating this loss in wellbeing indicated that parents with a chronically ill child would need 4275 euro a month to report the same well-being level as parents without a chronically ill child. For parents of children with a progressive illness (e.g. Duchenne and several metabolic disorders) the monetary value is 10925 euro, about 4.5 times their monthly income. The values for the ten different diagnoses studied, varied between 6959 to 10629 euro.
**Table 1 Overview of studies presented in this thesis**

<table>
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<tr>
<th>Chapter</th>
<th>Aim</th>
<th>Inclusion</th>
<th>Measurement</th>
<th>Outcome</th>
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<tr>
<td><strong>Chapter 2</strong></td>
<td>Overview of caregiver HRQoL and comparison group</td>
<td>n = 533 parents of children with chronic disease¹</td>
<td>Health Related Quality of Life in the past month</td>
<td>Parents of chronically ill children have a lower HRQoL. 50% at risk for HRQoL impairment.</td>
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<tr>
<td><strong>Chapter 3</strong></td>
<td>Exploring conceptual model of parental HRQoL</td>
<td>n = 543 parents of children with chronic disease</td>
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<td>Direct: female gender (-), parental age (+) chronic illness of the parent (-), care dependency (-). Mediator: holiday (+) and emotional support (+)</td>
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<td><strong>Chapter 4</strong></td>
<td>Employment and leisure activity time of parents and comparison group</td>
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<td>Employment (hrs/week) Leisure activity time (hrs/week)</td>
<td>Families with ill child work less hours a week: mothers work fewer hours. Also fewer hours leisure time when child is ill.</td>
<td>Salient factors: Being parent of child with chronic disease (-); use of child care (+); care dependency (-); low educational level (-)</td>
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<td><strong>Chapter 5</strong></td>
<td>Quantification of subjective costs of care</td>
<td>n = 580 parents of children with chronic disease n = 443 parents of schoolchildren</td>
<td>General well-being</td>
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<td><strong>Chapter 6</strong></td>
<td>Disease specific: HRQoL of mothers of children with sickle cell disease</td>
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<td>Health Related Quality of Life in the past month</td>
<td>Mothers of children with SCD lower socio-economic situation. SES-comparison: lower HRQoL on depressive moods, daily activities, vitality.</td>
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<tr>
<td><strong>Chapter 7</strong></td>
<td>Disease specific: Predictors of HRQoL of parents of children with metabolic disorders</td>
<td>n = 121 parents of children with metabolic disorders</td>
<td>Health Related Quality of Life in the past month</td>
<td>These parents have lower HRQoL on majority of HRQoL scales</td>
<td>-</td>
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</table>

¹ Parents from children from 10 diagnosis groups (Asthma, survivors of brain tumor, diabetes, Duchenne muscular dystrophy, Down syndrome, end stage renal disease, metabolic diseases, profound complex handicaps, sickle cell diseases, spina bifida)

² Parents from 2 elementary schools and 1 high school whose children were not chronically ill (parent report)
The overall conclusion of this thesis is that parents of chronically ill children were at risk for a lower quality of life, had a lower subjective well-being, families worked less hours a week in paid employment, fewer mothers took part in the labour force, and parents spent less time doing leisure activities than parents of healthy schoolchildren. In short, being a parent of a chronically ill child had consequences on physical, emotional, and social aspects of life. Salient factors negatively associated with these consequences were higher care dependency of the child, less emotional support, the parent being chronically ill, lower educational level, born outside the Netherlands and female gender. These results underline the importance of early detection of parents at risk and of parental support in clinical practice.

2 REFLECTIONS ON THE MAIN FINDINGS

2.1 A variety of consequences and potential impact on daily life
When confronted with a chronic illness, families have to deal with multiple demands simultaneously. Besides the usual family stressors, parents additionally have to deal with disease related stressors. Results of the present thesis show that being a parent of a chronically ill child resulted in adjustment of time doing ‘regular’ daily activities. Parents adjusted their paid work situation and leisure time. What’s more, parents evaluated their social, physical and emotional functioning more negatively than parents of healthy children, expressed in a lower HRQoL and subjective well-being.

The lower HRQoL reported by parents of chronically ill children is consistent with the literature. Quality of life of parents is often lower than among parents of healthy children.\textsuperscript{1-4} The meaning of a lower HRQoL is not easily described in clinically relevant levels of distress because there is no gold standard for a high and low HRQoL. We distinguished parents at risk for HRQoL impairment from parents not at risk, based on the 25\textsuperscript{th} percentile scores of parents of healthy children.\textsuperscript{5} This showed that almost half of the parents had scores below the threshold. The impact of being at risk on every day life is best explained by an example from the questionnaire used: “In the past month, did you have difficulties doing work, education or other daily activities? How much did this bother you?”. A low score means the parent reports an increase of problems and also negatively evaluates these problems. Overall, parents have a lower HRQoL with regard to cognitive functioning, sleep, pain, social functioning, performing daily activities, sexuality, vitality, positive and depressive emotions. These outcomes show that parents are bothered on all these parts of life which can be very disturbing in daily life.

Parents of children with a chronic disease may not be the most reliable employees. For instance hospital admission of the child and unexpected illness flares may interfere with the demands of paid employment. The solution several parents end up with, either or not by choice, is reduction of working hours of one of the parents, most often the mother (this thesis).\textsuperscript{6,7} The impact of these adjustments in employment on family functioning is easy to identify with, in light of the consequences of the financial crisis that started in 2008. Parents having difficulties meeting their job demands presumably are the first to lose their jobs when
the socio-economic situation deteriorates. Not being able to perform your job, or having to reduce working hours will result in deterioration of finances.8-10 And on the long run, might reduce chances of returning to the labour market. The emotional consequences of being (un)employed should not be neglected, because employment may contribute to mental health and personal development.11,12 One of the benefits of the reduced working hours is the extra time available to care for the child and their siblings and for instance, to spend time doing the large amount of administrative chores related to the child’s illness.

Parents report fewer hours of leisure activities per week than parents of healthy children. Conversations with parents revealed that parents adjusted their leisure activities (due to: e.g. lack of time and energy, difficulties with transportation, and misunderstanding from friends about the choices parents make) to better suit them to the family demands. Parents participating in sports competition (e.g. soccer or tennis) presumably experience problems meeting their team’s demands. And parents of a child with severe asthma who only take their child to smoke-free birthdays or parties, are often misunderstood by others and perceived as overprotective.13 Parents’ stress levels are higher while they have less time to relax, resulting in a disturbed balance. Being able to spend time doing leisure activities, meeting other people and to have contact with a social network is related to well-being, mental and physical health14-16 and is therefore of great importance.

All these consequences for parents may influence family functioning, and when placed together, the enormous significance and influence on the lives of parents becomes evident, as well as the accompanying subjective costs. The separate effects found in the Care-project are small to moderate. However, the total effect is large due to the number of parents experiencing these consequences and the multiplicity of consequences experienced.

2.2 Parents at risk
In general, being a parent of a child with a chronic disease is a risk factor for experiencing negative consequences on HRQoL, employment and leisure activity time. However, within group differences induce considerable variation in the consequences experienced by individual parents. Most vulnerable are parents with a low educational level, who are born outside the Netherlands, parents who are chronically ill themselves, and parents whose child had more functional limitations.

2.2.1 Socio-demographic characteristics
A low educational level, being female, being born outside the Netherlands, and being chronically ill as a parent are the main socio-demographic risk factors found in the Care-project. Protective factors were an older age and having a partner. Within literature, the role of socio-economic status (SES) on parental well-being is unclear. Several studies have found this association17-19, while other studies have not.20 However, comparison of results about SES is difficult, due to measurement problems.21 The concept SES is rather difficult to measure and proxy measurement is most often used. 21 Within the Care-project, inferences made, based on the proxy measures used (e.g. educational level and household income), imply
that parents belonging to a lower socio-economic group, who also have a child with a chronic disease are more vulnerable for a lower HRQoL, fewer hours employment and leisure activity time. No mediating effect was found for income on HRQoL. Perhaps not income, but financial concerns may explain HRQoL better. Parents have a lower household income and additionally presumably also have higher expenses due their child’s illness, leading to a double disadvantage and perhaps financial concerns.

Fourteen percent of parents in our sample reported to have a chronic illness, unfortunately no further description of disease characteristics is available. Parents who are chronically ill themselves appear to be more vulnerable for negative outcomes on HRQoL, well-being, employment, and leisure activity time. Although this result is not surprising, little research has been conducted on the topic of being a chronically ill parent with a chronically ill child. Most research has been performed among parents with chronic pain and multiple sclerosis, addressing psychosocial outcome in children. Chronically ill adults report problems with employment and a lower quality of life. Adding the consequences of having a child with a chronic disease to these results may explain the added vulnerability of this group. In addition, in case of heritability of the disease, we hypothesize that parents may experience feelings of guilt (also for parents who had been carrier of a defect) and may feel more responsible for the child.

In the Netherlands, in 2009 approximately 1 in 5 parents was a single parent (CBS), a number expected to grow in the near future. For single parents, the combination of care, employment and family life may be more challenging than for other parents. Within the Care-project, little evidence for this hypothesis was found. Single parenthood was not associated with employment and leisure activity time. Having a partner nonetheless does contribute to parental functioning because it is an important source of social support and thus contributes indirectly to HRQoL in our model. A possible explanation is that single parents have to work because of the financial consequences of being unemployed. Selection bias could also have caused these results, if only single parents participated who manage the consequences of having a chronically ill child well, and have enough support to combine work and care.

Gender differences in the experience of parenting a child with a chronic disease are evident. Mothers generally organize their life around the child (this thesis), while fathers are financial providers. Mothers on average report more emotional distress than fathers, while fathers report more finance and capacity related stressors. In addition, different factors may predict adjustment in mothers and fathers. Fathers tend to use more cognitive strategies in dealing with stress, mothers use more emotional strategies as seeking emotional support, and mothers and fathers are equally likely to search for information. Thus although in general being a parent of a child with a chronic disease is a risk factor, mothers and fathers differ in the consequences they experience.

2.2.2 Disease characteristics: care-dependency & disease development
The results of the Care-project demonstrate that care dependency of the child predicted HRQoL and hours of employment of the parents. Care dependency was defined as the sum of dependency of the child on the following aspects of daily life: physical care, mobility, eating
and drinking, medication use, coping with devices, amusing him/herself, contact with other children, and education. A higher score indicated a higher level of dependency on others. Our concept named care-dependency is best compared with the concept of functional limitation often used in the literature. Functional limitations of the child are known to influence parental functioning.\textsuperscript{1,32,33} A higher intensity of care is associated with more health and emotional problems in caregivers.\textsuperscript{34} In addition, although only the direct association between dependency on daily care and caregiver HRQoL was assessed, other factors may possibly moderate this relation. This is illustrated by the relationship between the parent and child. A better relation between parent and child leads to lower perceived burden in parents\textsuperscript{35} and to fewer behavioural problems in children. The quality of the relation between parent and child was not measured in this study, but should be considered in future research.

Besides the effect of care-dependency, progressive disease development in the past year, as reported by parents, was negatively associated with parental outcomes on HRQoL (metabolic disorders) and well-being. Both progressive and relapsing course of disease were expected to have the most consequences on parental daily life, as opposed to stable and improving course of disease. While each category poses specific demands on parents, relapsing and progressive illness demand continuous adaptation and add uncertainty, which may result in higher overall burden. Therefore, these parents may need continuous flexibility from their surroundings, and may for instance not be the most stable employee or friend. Contrary to our expectations, a progressive course of disease had no effect on employment situation. However, as it did have an effect on HRQoL (sleep, vitality) in parents of children with metabolic disorders, and on well-being in the total sample, progressive illness in the child is related to higher perceived burden in parents.\textsuperscript{2}

2.2.3 Protective factors
Besides risk factors, a number of protective factors has also been found. In addition to the already mentioned older age and having a partner, going on a holiday is beneficial for parents’ HRQoL, and parents receiving more emotional support have a lowered risk of HRQoL impairment. The beneficial effect of going on a holiday appeals to common sense, but is not often studied. The importance of social support for caregivers, on the other hand, has been acknowledged in many studies.\textsuperscript{2,17,18,36,37} In the Care-project, availability of emotional support served as a mediator between on the one hand socio-demographic and disease related variables and HRQoL on the other. Main effects of emotional support on HRQoL were also found, as well as a negative association of loss of friends with HRQoL in the group of parents of children with a metabolic disease. In a review by Lim & Zebrack\textsuperscript{4} both direct and mediating effects were found for social support on caregiver outcomes. Raina and others\textsuperscript{20} have only found an effect of social support on parental health, mediated by family function. They conclude that support from the direct family members appears more important than support provided by others. For parents of children with a chronic disease, support may have multiple direct and indirect effects. Emotional support contributes to quality of life and is mainly sought after by mothers\textsuperscript{31}, also instrumental support can help parents fulfil all their roles and therefore reduce stress levels or caregiver burden.
2.4 Caregiving process over time
Caregiving demands may differ across stages of the child’s disease and across developmental stages of the child. Depending on the course of the child’s disease, parents continuously have to adapt to a new status quo. In our study small effects of time on parental outcomes were found. A longer time since diagnosis led to more leisure activity time, older parental age was associated with higher monthly income, higher mental HRQoL (chapter 3), more leisure activity time, and higher maternal labour force participation (chapter 4). Older age of the child was positively associated with family employment (chapter 4). However, adding disease characteristics reduced the effect of older parental age on leisure activity time and maternal labour force participation. Also, older parental age was associated with lower vitality in parents of children with metabolic disorders (chapter 7). These results implicate an adaptation effect over time as parental HRQoL and participation in employment and leisure activity time increases. However, the results also show the influence of disease characteristics. The effect of functional limitation of the child and progressive course of the disease diminish the positive effect of time on parental functioning. These results suggest that parental HRQoL and participation may indeed increase in time when the child’s illness is not progressive and the child does not have severe functional limitations. An alternative explanation for these results is that the care giving tasks diminish because the adolescents who are chronically ill become more self-supporting, and the other children in the household also may need less attention. If this explanation is true, the effect of caregiver adaptation over time is not clear. Furthermore, age of the parent increases the chance that parents have to care for their parents. The additional burden of caring for both child and parent is a possible confounder influencing the effect of age on adaptation, although the number of parents caring for a parent is not expected to differ between the parents of chronically ill children and the comparison group.

Although the factor time was included in several analyses, based on our results no conclusions can be drawn about the influence of the phase of the disease or development on parental HRQoL and other outcomes. Parents included in the Care-project had their child’s diagnosis at least one year before taking part. This inclusion criterion was chosen in order to give parents time to come over the first shock of the diagnosis. Nevertheless, it would be interesting to gain more knowledge about the stages parents go through, following the model of medical traumatic stress developed by Kazak.

2.5 Generic and disease specific approach
Within the Care-project, a generic approach was adopted, assuming that chronic diseases in childhood share similarities and that these similarities are more important in predicting adaptation in parents than disease specific elements. Stein & Jessop were one of the first arguing the appropriateness of a division based on diagnosis. This approach has been adopted by Wallander and Varni in their well-known model of child and family adjustment. According to Stein & Jessop and Rolland, diseases may be similar in life risk perspective, impact of treatment, and functional disability. Several studies support this non-categorical
approach, although other studies suggest that unique disease characteristics influence parental adjustment.\textsuperscript{19,44} In the Care-project, 10 different disease groups participated. The aim was to include diseases varying in course of disease (e.g. stable versus progressive) and diseases with considerable and with little within group variation (e.g. asthma, Duchenne). In order to estimate disease characteristics, and because a general severity measure was not available, Care-dependency and course of the disease in the past year were used as measures of the impact of the ‘severity of the disease’. The main outcomes were analysed according to a generic approach. In addition, few disease specific effects of HRQoL and on well-being were investigated. The disease specific effects on well-being for instance, resemble the results based on course of disease. Effects were stronger for Duchenne muscular dystrophy and Metabolic diseases. Duchenne is a progressive disease, and within the group of metabolic diseases, 32\% of the children had a progressive course of the disease. The total HRQoL data showed overall lower scores, with strikingly lower scores for parents of children with metabolic disorders. Notwithstanding these results, evaluation of possible disease related predictors within the subgroup of parents (\textit{chapter 7}) did not reveal evident disease specific effects. Emotional support, progressive course of disease, and loss of friendship were the most evident predictors of parental HRQoL in this subgroup. Perhaps inclusion of disease specific parameters would have had an effect on parental HRQoL, however other studies reveal a minority of disease specific determinants, but more psychological determinants of parental outcome\textsuperscript{4,37,45-47}, concluding that a generic approach seems valid.

\textbf{2.6 Strengths & Limitations}

The care-study elaborates on the current literature by integrating the consequences of being a parent of a child with a chronic disease on employment, leisure activity time and HRQoL in one conceptual model. The use of a uniform measurement at one time point, including 10 diagnosis groups and a comparison group, based on a consensus based definition of chronic childhood disease, with age of the children ranging from 1-19 years old, led to a comprehensive set of information of a relatively large sample of parents. Another strength is the use of a generic HRQoL measure that included an evaluation of the physical, social and emotional health of the parents. This adds to current literature because it gives insight in how parents experience the ‘objective’ consequences on their physical, social and emotional functioning. The results of the care-project should be considered in light of a number of limitations. First, using a mailed questionnaire probably has resulted in non-response bias. The average response rate was 52\%. Length of the questionnaire and problems reading or writing may have decreased willingness to participate, and not all parents may be used to completing questionnaires.\textsuperscript{48} To increase response, active help with completion of the questionnaire was offered to parents who had difficulties reading and/or writing in Dutch or English, and all parents were phoned or received a reminding letter several weeks after the questionnaire was sent. No general characteristics of non-responders have been found across studies.\textsuperscript{49} Based on the characteristics of the participants of the Care-study and the patient populations, we assume that the non-responders were more often born outside the Netherlands,
had lower education, were more often a single parent, and were male. A low educational level is a risk factor for family employment (this thesis). Thus, if our assumption is correct, our results show an under representation of employment related problems. Also it may show an overrepresentation of HRQoL scores as women tend to present lower scores. However, the comparison of HRQoL scores in chapter 2 included correction for age and educational level of the parent, and in the regression analyses regarding employment these demographic variables were also accounted for (chapter 4). Though, in future research fathers should be approached more actively, and parents born outside the Netherlands could be actively approached by an informant from their own cultural background, using a method suitable to the population (e.g. interview).

A methodological limitation concerns the questionnaire used. It was developed for the Care-project and included two validated instruments, with adequate psychometric properties, as well as a number of questions adapted from other questionnaires, and questions developed by the researchers. For these questions, no information regarding reliability and validity was available. The clarity and applicability of the questions was pre-tested by 15 parents who met the inclusion criteria, and based on their comments, the questionnaire was adjusted. Also, the instruments were administered among all parents, also when they were not born in the Netherlands (18%). We used the TAAQoL for measurement of HRQoL. This instrument was developed in a well educated, non-minority sample, and we do not know whether the instrument is culturally sensitive and measures HRQoL adequately in other cultural samples. Another limitation of the TAAQoL is the lack of information about the clinical significance of the scores. It does not measure clinical depression, but merely the presence and evaluation of depressive emotions.

The use of a self-report measure and a single informant increased the possibility of finding relations that are in fact the result of shared method variance, meaning that the association between constructs may be due to the method used. In future research this problem could be solved by using a combination of for instance self-report and observations and multiple informants. Also, the cross-sectional approach did not allow inferences on causality of associations. However, in addition to straightforward regression analyses, we did analyse the direction of associations within the research model, using structural equation modelling. Still, the large number of variables included, the relatively small number of respondents, the size of the effects and the lack of longitudinal data do not allow confirmation of causality assumptions tested in the model.

The percentage of explained variance of the tested research model (chapter 3) was rather low. Inclusion of more psychosocial and behavioural determinants could contribute to the explanation of parental HRQoL. Factors not included in the Care-project were for instance coping style, personality, and communication between family members. Moreover, the occurrence of behavioural problems in children with chronic disorders is higher than among their healthy peers. Despite the fact that these behavioural problems may influence parental health, it was not measured in the present study as well as parental psychopathology. Depression is more often signalled in parents of chronically ill children, as well as traumatic stress, both concepts that may influence daily functioning. Future research should also address these topics.
3 CLINICAL IMPLICATIONS

Our data show that several parents experience a multitude of consequences, which has implications for clinical practice. Implementation of these clinical implications should be considered on several levels of society. A straightforward level is that of the children’s hospitals and pediatric hospital wards. Awareness of parental functioning is the key element that should be incorporated in the attitude of pediatricians and nurses. Furthermore, what society can do, is to be more flexible towards these parents. Being part of society, we can also do something ourselves by providing support to caregivers in our direct surroundings. Another level is that of policy makers who should be more aware of the complexity of the situation of the caregiver and the accompanying subjective costs.

3.1 Focus on the entire family in clinical setting
The focus of pediatric medical treatment used to be on the illness of the child. Nowadays, this focus is expanding towards a holistic approach of treatment outcome, including children’s physical, cognitive, and psychosocial development. Based on our results, parental functioning should be added as a parameter of treatment outcome in clinical settings. Parents experience several social consequences and have a lower HRQoL. Helping parents to minimize these consequences may help them to fulfil the different roles they perform. This would also contribute to treatment outcome in their children, the main goal of the pediatrician. And although not addressed in the care project, siblings also experience several consequences and should not be forgotten. The interaction between parents and their children was not addressed in the Care-project, but is known from the literature to influence both child and parent behaviour and should be integrated in family research. Therefore, family functioning should be incorporated as an important part of pediatric illness and its treatment.

3.2 Screening of parents at risk
First, all parents who are vulnerable for emotional, physical and social impairment should be recognized within the health care system and be given support if needed. In the Netherlands, general practitioners, home carers and pediatricians have most contact with parents of children with chronic diseases. Awareness and prevention are the main messages. Awareness of the possible consequences parents experience and how this may affect their children, treatment and family life. This should result in prevention of the deterioration of parent and family well-being. Recognition of parents needing help may be achieved by screening all parents of children with a chronic disease. However, screening of parents during a control visit for the child may be difficult to realize due to the limited time available. Within the Emma Children’s Hospital an intervention study on using patient reported outcome (PRO) in clinical practice is being prepared, including an online screening instrument for parents/caregivers (Parent Reported Outcome). Structurally monitoring the parents gives the possibility to immediately provide support when parents start reporting problems.
reduce time constraints, not pediatricians but social workers will receive these results and contact parents with alarming outcomes. Application of online screening both at home or in the waiting room is increasingly applied in disease populations, and has the future also for caregivers. Advantages are the large number of parents that can easily be included, the low costs and the great value of this information for clinical and research purposes.

3.3 Empowering parents
Interventions aimed at helping parents fulfilling all roles may be helpful for parents and will have a preventive function. Caring is a multifaceted and ongoing process, and parents need to keep everyone and everything together, (e.g. their child’s health, family life, school, work, hospital staff) which may require considerable energy and negotiation skills in the case of conflict of interest. Empowerment interventions aim to enable parents making sound decisions and to act according to their preference. Interventions aimed at parental empowerment have proven their benefit both for the children’s health and parental well-being.\textsuperscript{58-62} Parents have more feelings of control over their life, and the interventions lead to increased self-efficacy and self-awareness\textsuperscript{63} and fewer child adjustment problems.\textsuperscript{58} Interventions aimed at helping parents cope with their child’s disease by teaching skills and increasing their feelings of competence should therefore be implemented in clinical practice and research. This could also help parents to anticipate on their future needs and organize their lives according to these needs. Using the internet for online interventions should be seriously considered to increase the possibility of participation for parents. Furthermore, e-mental health applications for children with a chronic disease are also important. If children receive adequate care at home, this may interfere less with their parents live than an intervention at the hospital.

3.4 Stimulating respite care, holiday and leisure time
Parents should be stimulated to spend time doing things they enjoy. Handing over the care temporarily gives primary caregivers a break and provides relief.\textsuperscript{64-67} Also, engaging in sports or social activities contributes to better mental health.\textsuperscript{68-70} However, parents do not easily get, or take a break from care giving. It means handing over care giving tasks, which may be difficult to arrange or may emotionally be difficult. Fortunately, there are organisations dedicated at providing specialized home care to children. Health care providers could intervene between these organizations and parents. Parents should be stimulated to go on holiday with the entire family or alone. A positive effect of going on a holiday on parental HRQoL was found in this study. Parents often are unaware of the possibilities to either go on a holiday with the entire family, or to give their chronically ill child the possibility to go on a holiday camp. Several holiday accommodations exist for families with a family member needing (extensive) medical care. For instance the well-known Ronald McDonald house charities have two holiday accommodations in the Netherlands and organize several leisure time activities for children. Effort should be put in the development of easily accessible information regarding holiday options for these families.