

Development of a Quality of Life Instrument for Children with a Chronic Illness



Jeanet Bruil

**Health Psychology
Leiden University, The Netherlands**

**DEVELOPMENT OF A QUALITY OF LIFE INSTRUMENT
FOR CHILDREN WITH A CHRONIC ILLNESS**

PROEFSCHRIFT

ter verkrijging van
de graad van Doctor aan de Universiteit Leiden,
op gezag van de Rector Magnificus Dr. W.A. Wagenaar,
hoogleraar in de faculteit der Sociale Wetenschappen,
volgens besluit van het College voor Promoties
te verdedigen op donderdag 16 september 1999
te klokke 16.15 uur

door

Jeanet Bruil

geboren te Gorssel in 1963

Promotiecommissie

Promotor: Prof. Dr. C.M.J.G. Maes

Referent: Dr. T.M.T. van Elderen

Overige leden: Prof. dr. I.A. van Berckelaer-Onnes
Prof. dr. M. Boekaerts
Prof. dr. L.J.T. van der Kamp
Dr. J.M. Koot (Erasmus Universiteit Rotterdam)
Prof. dr. A.C.B. Peters (Universiteit Utrecht)
Prof. dr. J.M. Wit

Behorend bij het proefschrift

"Development of a quality of life instrument for children with a chronic illness."

Jeanet Bruil, 16 september 1999.

Stellingen

1. Alle definities van kwaliteit van leven dienen zowel het functioneren van mensen als hun afweging van het belang daarvan te bevatten (dit proefschrift).
2. Kwaliteit van leven instrumenten voor kinderen kunnen slechts voor een beperkt leeftijdsbereik worden ontwikkeld (dit proefschrift).
3. Het oordeel van ouders over de kwaliteit van leven van hun kinderen is per definitie niet valide (dit proefschrift).
4. De veronderstelling dat kinderen met een chronische ziekte enkel negatieve gevoelens over hun ziekte hebben, zegt meer over de beleving van onderzoekers dan over die van kinderen (dit proefschrift).
5. Samenwerking in psychosociaal onderzoek in de kindergeneeskunde is essentieel, maar wordt tegengewerkt door eigenbelangen of instituutbelangen.
6. De stormachtige groei van prenatale screening wordt gevoed door de bedenkelijke gedachte dat een kind gezond is zolang er geen constructiefout is aangetoond.
7. Psychologen letten teveel op het gedrag in plaats van op de situatie terwijl voor pedagogen het omgekeerde geldt.
8. De psychologie zal meer aandacht moeten hebben voor wat mensen op de been houdt in plaats van wat hen ten gronde richt.
9. Het krijgen van kinderen tijdens een promotie-onderzoek bevordert de kwaliteit van leven van de promovenda maar ook de kwaliteit van haar proefschrift.
10. Alle AIO's dienen bij hun aanstelling een reserve-promotor te hebben.

..... de wereld is niet mooi, maar jij kan haar een beetje mooier kleuren ...
H. van Veen (1986)

Aan mijn ouders
Voor Ruud, Pepijn en Lars

Cover design: Hans de Wolf en Sander Spaargaren
Printed by Printpartners Ipskamp, Enschede

Bruil, Jeanet
Development of a quality of life instrument for children with a chronic illness.
Doctoral dissertation Leiden University - With ref. - With Summary in Dutch.
ISBN 90-9012920-0

Financial support for the printing of this doctoral dissertation has been kindly provided by the Faculty of Social Sciences, Leiden University and TNO Prevention and Health, Leiden.

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VOORWOORD

Het onderzoek dat in dit proefschrift beschreven is, startte bij de ervaringen van kinderen met een chronische ziekte en hun ouders. Ik dank de vele kinderen, kinderen met een chronische ziekte en gezonde kinderen, en hun ouders voor hun medewerking. De hartverwarmende opmerkingen van ouders waarin het belang van het vragen naar de kwaliteit van leven werd benadrukt, stimuleerden mij om door te gaan. Enorme bewondering heb ik voor de veerkracht van kinderen en ouders.

Zonder de deelnemende specialisten, behandelende artsen had ik de ouders niet kunnen benaderen. Hen wil ik danken voor de aandacht die zij, in hun drukke praktijk, aan het onderzoek hebben willen besteden. In het bijzonder dank ik het team van het Wilhelmina Kinderziekenhuis Utrecht voor hun samenwerking met betrekking tot het onderzoek bij de kinderen met epilepsie. Voorts dank ik het Jan van Breemen Instituut te Amsterdam voor de medewerking aan de ontwikkeling van het reuma-specifieke deel van de HAY. Cobi en Jos van het Rijnland Ziekenhuis Leiderdorp dank ik voor de medewerking aan het onderzoek bij kinderen met diabetes mellitus. Lisette en haar collega's van het EMGO-Instituut VU Amsterdam dank ik voor de samenwerking in de ontwikkeling van de astma-specifieke deel van de HAY.

Terugkijkend naar de start van het onderzoek, ben ik blij dat ik destijds heb doorgezet. Onze start was geen vliegende start Irma. We zijn door diepe dalen gegaan en hebben vaak getwijfeld of het er nog van zou komen. Ik ben blij dat je vandaag naast mij staat. Laat dit niet het einde zijn van onze betrokkenheid bij onderzoek naar kinderen met een chronische ziekte. Rita, Jacqueline, Anja, Thuly, Kirsten en Janneke dank ik voor de hulp bij de dataverzameling en de datainvoer. Elise, ik dank je voor de heldere commentaren op mijn analyses. Kate Hudson bedank ik voor de kritische en snelle correctie van het Engels. Ariena, ik dank je voor je hulp bij de voorbereiding van mijn promotie. Hans en Sander, dank voor de hulp bij het maken van het kافت.

Bij het COJ ontwikkelde ik mijn liefde voor onderzoek. Van jullie, Kees en Peter, heb ik veel geleerd. Margot, ik weet nog goed hoe wij samen onderzoeksproblemen oplosten middels heftige discussies. Mijn oud-collega's van Klinische en Gezondheidspsychologie, en in het bijzonder Margot, Winnie, Chris, Laura, Mike, Kate, Ivan, Hende, Anneloes en Hetty, wil ik bedanken voor hun interesse, hartelijkheid, hulp en geruststellingen. Ik kwam als mee-eter in de vakgroep, maar jullie hebben mij dit gevoel snel doen vergeten. Arja, thank you for your support and friendship. Your enthusiasm stimulates me to go on in this research area. Nadia, met jou heb ik helaas maar kort kunnen samenwerken. Mike, Erik-Jan en Marc, tijdens de

vergaderingen van de Computercommissie heb ik veel over de computertechniek geleerd.

Nog voor mijn proefschrift afgerond was, kwam ik in het warme nest van de Divisie Jeugd van TNO-PG. Erik, ik wil je danken voor de tijd die ik kreeg om mijn proefschrift af te ronden. De aanmoediging en hulp van jou, Ton, Marja, Mascha, Ineke, Minne, Miranda, Pauline en alle andere TNO-ers doen mij goed.

In de loop der jaren heb ik heel wat mensen verwaarloosd, familie en vrienden. Eerst het proefschrift en toen ook nog kinderen, lieten minder tijd over voor kroegen, uit eten gaan, sporten en andere leuke dingen. Ik hoop dat jullie het mij vergeven. Lisette, een paar jaar geleden maakte ik onwetend jouw stress mee. Ik vind het heel leuk dat jij vandaag nu naast mij staat. Hans en Riet, op jullie hulp varen wij. Papa, mama, jullie hebben mij altijd gesteund en zonder jullie motto: "waar je aan begint moet je ook afmaken", was ik misschien eerder gestopt. Lieve Ruud, het leven is zo druk, twee kleintjes en dan ook nog dat proefschrift. Reikhalzend werd vooral door jou uitgekeken naar het moment waarop het naar de drukker kon. Bedenk echter goed, de was vouwen blijft ook zonder proefschrift jouw taak. Jouw nuchtere commentaar en positieve levensinstelling helpen mij om te relativieren. Jouw nachtelijke uren aan de lay-out waren onmisbaar. Pepijn en Lars, jullie herinneren mij er elke dag aan dat er meer is dan werk alleen. Ik geniet iedere dag van jullie. Daarnaast besef ik des te meer, zeker door jullie ziekteperiodes, dat het belangrijk is om zowel in onderzoek als in de klinische praktijk kwaliteit van leven te betrekken.

INTRODUCTION AND OUTLINE

Children with a chronic illness have to cope with their illness during a long period or even for their lifetime. This will have a large impact on both the child and the family. Illness characteristics and limitations, but also medical intervention may impact negatively on the child's development. As a consequence, the attention of health care providers has widened from survival and biomedical status to the behavioral and psychosocial status, and a subsequent increase in attention devoted to pediatric or child health psychology has been reported (Johnson & Johnson, 1991).

Two basic approaches have been used to study the consequences of childhood chronic illness, the categorical and the noncategorical approach. The *categorical approach* groups chronic conditions in terms of specific diagnoses as asthma or diabetes mellitus (Stein & Jessop, 1989; Thompson & Gustafson, 1996). Such a condition-specific approach can facilitate the development of new and relevant scientific information and is compatible with the work of pediatric colleagues. On the other hand, research according to the categorical approach is conducted among small groups so that results are not comparable, and interventions are only available for a minority. The categorical approach can present serious obstacles to the development of valid scientific knowledge that can be generalized to other chronic illnesses. The second approach is the *noncategorical approach* (Stein & Jessop, 1984, 1989), which assumes that children with a chronic illness have common life experiences and problems stemming from generic dimensions of the medical condition, such as onset, course, prognosis or paroxysmality. Currently the noncategorical approach is generally accepted. Nevertheless, each illness also has unique aspects that may influence the child's daily experiences (Nelms, 1989). The *modified categorical approach* is therefore preferred (Lavigne, Ross, Berry & Hayford, 1993; Thompson & Gustafson, 1996; Holden, Chmielewski, Nelson & Kager, 1997). In this approach, generic illness characteristics as well as unique features are incorporated. The modified approach is used in this doctoral dissertation to focus on generic as well as disease-specific features and consequences of childhood chronic illness.

Four illnesses are therefore selected in this doctoral dissertation, these being asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. These illnesses are similar in several dimensions resulting in common experiences that may influence the child's functioning. The illnesses are viewed as nonfatal, children with these illnesses are generally able to attend normal schools and to participate in social activities. Furthermore, the management requires daily treatment, diet or exercise. In addition, each illness also has unique aspects in paroxysmality, brain involvement and physical impairment that may influence a child's daily experiences and his or her functioning in normal daily life.

The consequences of illness are influenced by several mediators, such as disease characteristics, family variables and coping resources, and are noticed on the level of physical, social and psychological functioning of children. Knowledge about the consequences of a chronic illness on these dimensions is helpful when diagnosing children in need of intervention and when evaluating interventions. The current focus in research, as well as in clinical practice, is therefore to integrate the physical, social and psychological consequences of having a chronic illness. This means that not only physiological but also broader health-related outcomes are important, including psychosocial measures (Pantell & Lewis, 1987).

Health-related quality of life instruments can be used to evaluate the functioning of children in these three dimensions of functioning. The aims of using health-related quality of life measures include the evaluation of the efficacy of the medical interventions, the identification of secondary dysfunction or residual dysfunction of long-term survivors and the identification of subpopulations at risk for psychological or behavioral problems (Spieth & Harris, 1996).

Despite the fact that nearly 20% of children are chronically ill, attention for health-related quality of life among adults and the growing area of research in child health psychology, research on quality of life in pediatric care is, whilst increasing, as yet very limited. This is partly due to the fact that there is a shortage of suitable quality of life measures for children with a chronic illness (Mulhern, Fairclough, Friedman & Leigh, 1989; Rosenbaum, Cadman & Kirpalani, 1990; La Greca, 1994, Elderen et al., 1994). In response, the HOW ARE YOU? (HAY), a health-related quality of life questionnaire for children of 7 to 13 years with a chronic illness, including children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis, was developed. In this doctoral dissertation, the theoretical background, and the development, validity and reliability of the HOW ARE YOU? (HAY) are described.

In *Chapter 1*, the definition and prevalence of childhood chronic illnesses are described. In addition, a stress and coping model is used to describe important mediating variables in the adaptation to childhood chronic illness. The model is used as an introduction to the description of the consequences of chronic illness for the child's functioning.

In *Chapter 2*, an illustrative review of studies on the physical, social and psychological functioning of children with asthma, diabetes, epilepsy and juvenile chronic arthritis is provided.

In *Chapter 3*, the definition of health-related quality of life and its assessment among children are described. These three chapters together form the prelude to the subject of this doctoral dissertation: the development of a health-related quality of life questionnaire, the HOW ARE YOU? (HAY)

In *Chapter 4* the developmental stage and the pilot study of the HAY are described. Furthermore, the concurrence between child and parents' scores is discussed. Finally, the amendments made in the pilot version of the HAY are delineated.

In *Chapter 5* the results of the main study are described in which the validity and reliability of the HAY was tested using a large group of healthy children and children with a chronic illness. In addition, the concurrence between child and parent's reports is discussed.

In *Chapter 6* the data presented and its implications for use of the HAY are discussed.

CHAPTER 1: CHRONIC ILLNESS IN PRIMARY SCHOOL CHILDREN

1.1 A DEFINITION OF CHILDHOOD CHRONIC ILLNESS

Advances in medical knowledge and treatment procedures have resulted in effective treatments or even cures for many illnesses. Chronic illnesses, however, remain a challenge to medical science, for these illnesses are either debilitating, fatal or can be treated successfully but treatment is life-long. Although common characteristics exist, there are vast differences in course, changeability and consequences among chronic illnesses, so that a single, universally accepted medical definition of chronic illness does not exist (Maes, Leventhal, & de Ridder, 1996). The definition applied in this doctoral dissertation fits with most current descriptions and refers to "a physical condition that affects children for extended periods of time, often for life. The illnesses can be 'managed' to the extent that a degree of pain control or reduction in attacks (of asthma), bleeding episodes (in hemophilia) or seizures can generally be achieved. However, they cannot be cured." (Eiser, 1990b; p.3).

Most researchers define illnesses as chronic when they have an actual or expected duration of longer than three months. While somewhat arbitrary, Pless and Satterwhite (1975) have shown that the 3 months criterion correctly identified 91.8% of those judged independently as having a chronic illness and 81.8% of those ultimately judged to be healthy (Pless and Satterwhite in Perrin et al, 1993).

1.2 GENERAL OUTCOMES OF STUDIES ON THE PREVALENCE OF CHILDHOOD CHRONIC ILLNESS

Whilst the prevalence of some somatic chronic childhood illnesses has declined markedly, prevalence estimates on the whole are not decreasing. On the contrary, the prevalence of children with a chronic illness has doubled in the 20 year period from the early 1960s (Thompson & Gustafson, 1996). One reason for this is that many children with a chronic illness who previously would have died, now survive into adulthood (Newacheck & Taylor, 1992). For instance, with an illness like cystic fibrosis for which the age expectancy is enhanced but still severely curtailed, current survival rates indicate that most children with cystic fibrosis live into the third decade (Perrin & MacLean, 1988). Another cause for stability in prevalence estimates is the decline in mortality among very low birth weight infants, whilst the rate of moderate to severe handicaps in this group remains relatively stable (Thompson & Gustafson, 1996). A number of new conditions, such as AIDS and prenatal drug exposure, also contribute to the prevalence of chronic illnesses (Thompson & Gustafson, 1996). In addition, it has been suggested that the prevalence of non-life threatening chronic conditions may have increased in recent years (Newacheck & Taylor, 1992).

Studies vary in estimated prevalence of chronic illnesses in childhood because of differences between the samples used and the methods chosen. Some studies are population-based, some studies are clinic-based. Some studies rely on parent reports, others use medical records. It appears that parents tend to overestimate the prevalence of clinically diagnosed chronic conditions, but that the over-reporting declines with the severity or perceived stigma of the condition (Gortmaker, Walker, Weitzman & Sobol, 1990; Newacheck & Taylor, 1992). Interview methods can also alter the estimates, as estimates are higher when the respondent has to indicate on a preselected list whether he or she has the condition (Berg & Bos, 1989).

The epidemiology of childhood chronic illness differs from that of adults' chronic illness; children face a large number of rare illnesses, whilst adults face a much smaller number of common illnesses (Perrin & MacLean, 1988). A large number of rare illnesses leads to the fact that parents, children and the community at large suffer from a relative lack of experience and familiarity with these illnesses (Thomas & Gustafson, 1996), which may cause difficulties in managing the illness and rearing the child in family, school or society.

1.3 OUTCOMES OF NATIONAL HEALTH STUDIES IN THE US AND THE NETHERLANDS

A frequently cited epidemiology study is the ongoing nationwide National Health Interview Survey (NHIS) being held in the US. In 1988, a special supplementary questionnaire on child health was included in this survey which revealed that an estimate of 31% of children under 18 years of age had one or more chronic conditions (Newacheck, Halfon & Budetti, 1986; Newacheck & Taylor, 1992; Newacheck & Stoddard, 1994). The overall prevalence of chronic conditions was similar for adolescents and younger children, boys were more frequently reported as being chronically ill than girls (Newacheck & Taylor, 1992). When, according to the definition described above, only those conditions considered to have a duration of three months were included, the percentage of children considered to be chronically ill dropped from 31% to 19% (Newacheck & Stoddard, 1994). This is, however, an upperbound estimate because no severity data were used. Taking into consideration limitations and bother, 20% of the US children in 1988 experienced mild chronic conditions, 9% experienced chronic conditions of moderate severity, at least on the basis of physiological data, and only 2% to 4% experienced severe chronic conditions that affect their daily activities on a regular basis (Newacheck & Stoddard, 1994; Perrin & MacLean, 1988).

The diagnoses in the NHIS study differed substantially at each extreme of the severity spectrum. Children with mild chronic conditions were much more likely than children with severe chronic conditions to have respiratory allergies and repeated ear infections. In contrast, children in the severe group were more likely to

have muscular impairments, hearing and speech impairments, cerebral palsy, diabetes, asthma, epilepsy, and arthritis (Newacheck & Taylor, 1992). Respiratory illnesses (principally asthma) accounted for 41% of all activity limitations caused by chronic conditions and injuries (Newacheck, Halfon & Budetti, 1986). The data indicated that children experienced an added burden of illness through school absence or physician contacts (Newacheck & Taylor, 1992).

In the Netherlands, the Health Interview Survey of Statistics, a similar study, although smaller than the NHIS study in content and number of participants, has been carried out among families by the Central Statistical Office (CSO) annually since 1981. This survey included questions on the prevalence of 24 chronic conditions, which revealed that 18% of the children between 0 to 14 years of age had a chronic illness.

In the next paragraph estimates drawn from the CSO study and the NHIS study are provided.

1.4 PREVALENCE ESTIMATES OF CHILDHOOD CHRONIC ILLNESSES IN THE NETHERLANDS AND THE US

The prevalence estimates of childhood chronic illnesses in the Netherlands and the US are given below using the CSO study (Wulp, 1996) and the NHIS study of 1988 (Newacheck & Stoddard, 1994).

Table 1.1: Prevalence of children with one or more chronic illnesses in the Netherlands and the United States

	CENTRAL STATISTICAL OFFICE 1994/1995 ¹ 0-14 years (%)	NHIS 1988 ² 0-17 years (%)
Children with one chronic illness	15.0	14.6
Children with two chronic illnesses	2.4	3.5
Children with three chronic illnesses	0.3	
Children with three or more chronic illnesses		0.9
Children with four or more chronic illnesses	0.1	
The total percentage of children with one or more chronic illnesses	17.8	19.0

¹ Sample size 3,492 children of 0 to 14 years old based on 1994/1995 (Van der Wulp, 1996)

² Sample size 17,110 children under 18 years old based on 1988 (Newacheck & Stoddard, 1994)

As shown in Table 1.1, both national studies report an estimated prevalence of nearly 20%, the prevalence of children with only one chronic illness is 15%.

The prevalence estimates of the distinct illnesses however vary. In Table 1.2 the prevalence estimates from both studies for the distinct chronic illnesses are presented.

Cultural and racial differences between the Netherlands and the US make the difference in the prevalence of chronic illnesses comprehensible. As described above, the variation in estimates can also stem from using different sources, methods, definitions, so that comparisons are difficult to make. Both studies used the parental reports as sources of data. Differences could stem, however, from the methods used. In the US study, respondents were read a list of illnesses and asked to indicate whether the sample child had any of the included illnesses during the past 12 months. These illnesses were based on a selected list of illnesses prevalent in children. In contrast, the Central Statistical Office study used a limited list of 24 illnesses for children, which was originally drawn up for use among adults. The Dutch method may thus lead to an underestimation of prevalence, as there are certain illnesses that are typical of childhood that were not included in the questionnaire. As the Dutch situation is studied in this doctoral dissertation, this list was used in the table. As a consequence, the list of illnesses of the NHIS is not complete.

Table 1.2: Prevalence of distinct childhood chronic illnesses in the Netherlands and the United States

ILLNESS	CENTRAL STATISTICAL OFFICE	
	1994/1995 ¹ 0-14 years (%)	NHIS 1988 ² 0-17 years (%)
Chronic a-specific respiratory diseases (asthma, chronic bronchitis)	9.1	4.3
Perinatal-frontal or maxillary sinusitis	5.1	n.a.
Serious skin disease	1.8	n.a.
Eczema and skin allergies		3.3
Disorder of the large or small bowel for longer than 3 months	1.7	n.a.
Migraine	1.3	n.a.
Chronic spinal affections for longer than 3 months, slipped disc	0.6	n.a.
Chronic cystitis	0.4	n.a.
Epilepsy	0.3	n.a.
Epilepsy or convulsions without fever		0.2
Serious heart disease	0.2	1.5
Arthritis of hands or feet	0.2	n.a.
Other rheumatoid arthritis for longer than 3 months	0.1	n.a.
Arthritis or joint problems		0.5
Serious disease of the kidney	0.1	n.a.
Diabetes mellitus	0.1	0.1
Dizziness with falling	0.1	n.a.

Note. Diagnoses in the Central Statistical Office study have been used as selection criteria. [n.a. not available]

1. CSO, sample size 3,492 children 0-14 years 1994/1995 (Vademecum Gezondheidsstatistiek Nederland, 1996)
2. NHIS, 1994, based on 1988, sample size 17,110 children under 18 years old (Newacheck & Stoddard, 1994)
3. Illnesses that are only prevalent above 14 years of age as stroke, stomach ulcer were excluded

Another cause of variation in estimates is the definitions applied. One example is the definition for asthma. In the Dutch study, a chronic *a-specific* respiratory disease (in Dutch the abbreviation CARA is used) incorporates also hay fever and other respiratory ailments whereas in the NHIS study, these illnesses were included separately (Newacheck & Stoddard, 1994).

Although the studies are therefore difficult to compare, it can be generally concluded that both studies support the estimate of around 20% prevalence of chronic childhood illnesses and, in particular, a relatively high prevalence of children with respiratory illness.

1.5 PREVALENCE OF CHRONIC ILLNESSES AMONG BOYS AND GIRLS IN THE NETHERLANDS

The studies carried out between 1989 to 1993 by the Central Statistical Office provided separate estimates of chronic illnesses among boys and girls (Wulp, 1996). In 1.3 these prevalence estimates are presented.

Table 1.3: *Prevalence estimates of chronic illnesses among boys and girls, 5 to 15 years old, in the Netherlands*

	Total group %	Boys %	Girls %
Children with one chronic illness	11.9	12.7	11
Children with two chronic illnesses	1.9	1.9	1.9
Children with three or more chronic illnesses	0.3	0.4	0.2
Children with four or more chronic illnesses	0.0	0.0	0.0
The total percentage of children with one or more chronic illnesses	14	15	13

Sample size 5,533, boys 2,830, girls 2,703, CSO study 1989/1993 5 to 15 years (Wulp, 1996)

Overall, there is a slightly higher prevalence of chronic illness among boys than among girls (Van der Wulp, 1996; Newacheck & Stoddard, 1994). In Table 1.4 the estimated prevalence of distinct illnesses among boys and girls in the Netherlands is presented.

The chronic illnesses of greatest concern to the community are those that are the most prevalent, have a major impact on the health care system and/or have a high rate of mortality (Maes, Leventhal, & de Ridder, 1996). It is well known that asthma is the most prevalent chronic illness in this young age group. Malignant neoplasms are one of the major causes of death in this age group, if one excludes causes not related to chronic illness as injuries or poisoning which are the most frequent cause of death (Wulp, 1996). Whereas boys are, in general, more frequently ill than girls, prevalence

estimates show also specific gender differences for distinct illnesses. While asthma, for instance, is more prevalent among boys, chronic cystitis is more prevalent among girls.

Table 1.4: Prevalence of distinct chronic illnesses¹ among boys and girls, 5 to 15 years old, in the Netherlands²

Illness	Total group (%)	Boys (%)	Girls (%)
Chronic a-specific respiratory diseases (asthma, chronic bronchitis)	7	8.1	5.3
Perinatal-frontal or maxillary sinusitis	3.4	3.3	3.6
Migraine	1.4	1.8	1.1
Serious skin disease	1.4	1.6	1.1
Chronic spinal affections for longer than 3 months, slipped disc	0.7	0.9	0.9
Disorder of the large or small bowel for longer than 3 months	0.6	0.6	0.7
Epilepsy	0.5	0.6	0.5
Chronic cystitis	0.5	0.2	0.7
Serious heart disease	0.2	0.1	0.3
Dizziness with falling	0.2	0.1	0.6
Disease of the liver	0.1	0.1	0.1
Serious disease of the kidney	0.1	0.1	0.3
Diabetes mellitus	0.1	0.2	0.1
Arthritis of hands or feet	0.1	0.1	0.0
Other rheumatoid arthritis for longer than 3 months	0.1	0.0	0.2
Malignant neoplasm or cancer	0.1	0.0	0.1

1. Illnesses that are only prevalent above 14 years of age as stroke, stomach ulcer are excluded
2. Sample size 5,533, boys 2,830, girls 2,703, CSO 1989/1993 5-15 years (Wulp, 1996)

From the estimates mentioned above it can be concluded that one in every five children has a chronic illness in both the Netherlands and the US. The American NHIS study revealed that half of this group experiences moderate to severe limitations in daily life. This information, however, does not answer the question "What are the consequences for daily life for these children and their families?". Unfortunately there is no straightforward answer to this question. Many factors other than the specific diagnosis contribute to how children with a chronic illness and their families function.

1.6 IMPACT OF CHRONIC ILLNESS ON CHILD AND FAMILY

Chronic or long-term childhood illness affects the functioning of both child and family. The presence of a chronic illness creates special tasks and problems that may interfere with the child's normal growth and physical, social, and psychological development. There is ongoing need for medical care, special education services and a range of social services. Daily treatment regimens and the management of daily activities require extra time and skill of both parents and children. In addition, the way others respond can create an additional emotional strain.

There are, however, contrasting findings regarding the psychosocial concomitants of physical disorders due to the lack of reliable instruments, the use of different

perspectives, use of small samples and the fact that different chronic disorders have been investigated in the studies (Wallander, Varni, Babani & Banis, 1988).

Studies emphasize the variability in outcomes and suggest that there are numerous factors that interact in complex ways to determine the adaptation of the child. Although children with a chronic illness were previously compared with healthy children to understand the deviation from the norm, the current view is that the psychosocial problems these children may experience can be best understood as normal reactions to real life stressors, which casts the children in a normalizing framework (Perrin & Mac Lean, 1988; Eiser, 1990a).

To understand fully the impact of a chronic illness on child and family, it is essential to consider it in the context of illness characteristics, child development, family life, and psychosocial variables. Although a large number of studies included a selection of risk and resistance factors and outcome factors, the theoretical framework of most of the studies is still rudimentary (Lemanek, 1994). Research in the past was hampered by a lack of theoretical or conceptual frameworks, which are necessary to guide research studies, integrate findings and implement interventions (Thompson & Gustafson, 1996).

Nowadays several models are available to guide research in pediatrics and are being applied more often. Although it is beyond the scope of this doctoral dissertation to provide an exhaustive review of stress and coping models, main characteristics of a few of the models that are available will be described.

One model is described by Rolland (1987) and tries to connect illness with individual and family development. The main focus in this model is placed on a psychosocial typology of illness and time phases of illness development. A broad range of illnesses is grouped according to the key biological similarities and differences that dictate distinct, psychosocial demands for the child and family. These include the category of onset (acute or gradual), the category of course (progressive, constant or relapsing), the category of outcome and the category of the degree of incapacitation. Secondly, the family's history and coping with illness is related to these illness characteristics and together they relate to the adaptation to a chronic illness. Thirdly, the illness, individual and family life cycles are recognized as interwoven threads. Rolland highlights herewith two important issues in his model: the importance of general illness characteristics and the importance of development in child and family life cycle.

A second model worth mentioning has often been used among children with a chronic illness to study their adaptation and is described by Wallander and Varni (1989; 1992). This model is based on research among children and the theoretical developments put forth by Pless and Pinkerton (1975). The conceptual emphasis is placed on the stress the children experience and on the coping resources that may be available to them in

dealing with this stress. Examples of risk factors include illness parameters and psychosocial stressors, resistance factors include family resources and coping strategies (Wallander, Varni, Babani, Banis & Wilcox, 1989; Wallander & Varni, 1992). The third model was developed by Perrez and Reicherts (1992) who enlarged the stress-coping model of Lazarus and Folkman (1984) by adding valence, controllability, changeability, ambiguity, and recurrence as key dimensions of a situation. In this approach, the key dimensions by which the illness is characterized are important, not the medical diagnosis.

The model used in this doctoral dissertation is described by Maes et al (Maes, 1993; Maes, Leventhal & de Ridder, 1996) and is also based on the stress-coping model of Lazarus and Folkman (1984). The basic assumption of this model is that people who are confronted with a stressor, such as a childhood chronic illness, evaluate this stressor and that this evaluation then determines the emotional or behavioral reactions. The model was extended by illness characteristics (see also Rolland, 1987; Perrez & Reicherts, 1992, Stein & Jessop, 1989). In addition, other (stressful) life events were added with their influence on coping. Finally, life goals or values and social relationships were added influencing the meaning of the illness and the selection of coping (Maes, Leventhal & de Ridder, 1996). Although this stress and coping model was developed for studying adaptation among adults with a chronic illness, this model is used as a guideline to describe some important contributions to the knowledge of the determinants of the impact of childhood chronic illness.

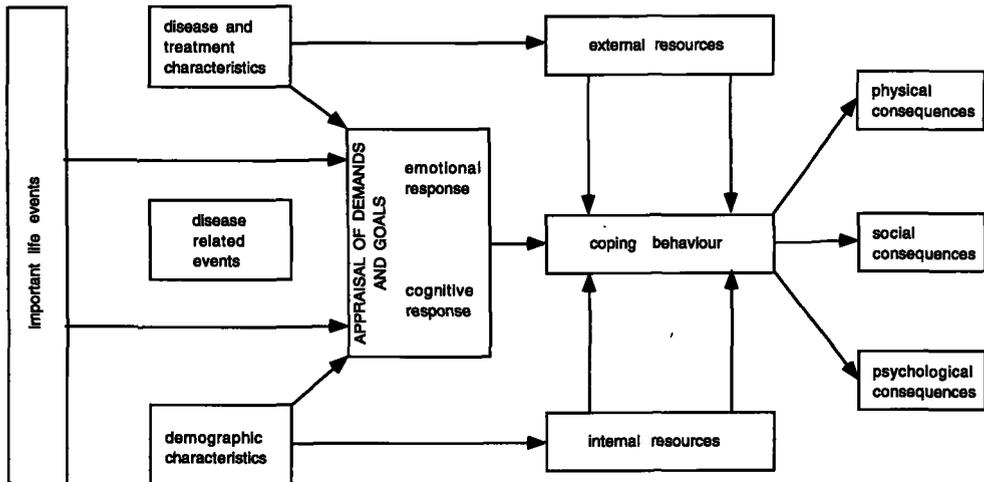
1.7 COPING WITH A CHRONIC ILLNESS, A THEORETICAL FRAMEWORK.

In the following a review of studies on risk and resistance factors and consequences in the child's adaptation to chronic illness are provided, using the model of Maes, Leventhal and de Ridder (1996) as a theoretical framework. The following describes the model when used in pediatrics (see Figure 1.1).

According to this model, *other important life events* can influence the child's appraisal as well as the family's appraisal of a chronic illness. Divorce of the parents, a parent losing a job, illness or death of other family members will be important factors in the appraisal of illness although not studied specifically in children. In addition, there are critical transitions in the child's development and family life, which will influence the appraisal of illness and necessitate reappraisal (Rolland, 1987). Children may go to a day care center when they are very young or go to school at the age of four, as a result of which the daily care of the child falls not only on the parents but also on teachers. With increasing age, children and family have to cope with the additional strain caused by changing developmental and social tasks. The change to secondary school around the age of twelve is, for instance, a normal life event that can cause stress in every child and family. Children with a chronic illness

however have an added stress factor because they also have to take their illness into account. The number of teachers is increased, school day hours and daily life rhythm is changed so that often the self-management tasks are more difficult to fit in and the child itself has to assume responsibility in these matters. Often the school is further from home and parents will not be readily available.

Figure 1.1: Coping with childhood chronic illness (Maes, Leventhal & De Ridder, 1996)



Chronic illnesses differ in terms of *disease and treatment characteristics*, the characteristics previously described by Rolland (1987) and Perrez and Reicherts (1992) come to mind. In studies among children with a chronic illness, two classification categories of disease characteristics are often used. The first one is the categorical approach, which groups chronic illness in terms of specific diagnoses as asthma or diabetes mellitus (Stein & Jessop, 1989; Thompson & Gustafson, 1996). The second approach is the noncategorical approach (Stein & Jessop, 1989), which assumes that children with a chronic illness have common life experiences and problems stemming from generic dimensions of the medical illness. Currently the modified categorical approach is preferred, as this approach includes generic dimensions as well as unique aspects of illness and treatment (Thompson & Gustafson, 1996; Lavigne & Faier-Routman, 1993; Holden, Chmielewski, Nelson & Kager, 1997). In this approach, generic illness characteristics including onset, course, prognosis, but also brain involvement, visibility, as well as unique features, such as breathlessness in asthma, are considered to be important.

The onset can be divided into acute, such as stroke, or gradual such as in cystic fibrosis. In an acute illness, family and child have a very short period to adjust. The rate of change required within the family to cope with a gradual-onset illness is less

and enables a more protracted period of adjustment (Thomas & Gustafson, 1996). The course of illness can be constant as in cerebral palsy or episodic as in epilepsy. Episodes may cause periods of increased stress and uncertainty because of their unexpected nature as in asthma or epilepsy. Despite increases in life expectancy for many formerly fatal illnesses, some illnesses are still fatal or incorporate the threat of being fatal. The awareness of the threatening nature of illness as in cancer is apparent even after cessation of treatment (Dongen-Melman, 1995).

Illness in which there is brain involvement appear to have an increased impact on the family (MacLean, 1988). The degree of visibility has also been associated with the degree of problems, possibly because of the stigma and the social isolation it causes. On the other hand, it has also been reported that visibility might be a protective factor because there is no need to think about telling others (Zimmerman, 1995; Perrin, Ayoub & Willet, 1993; Jessop & Stein, 1985). It has been suggested that invisibility of the illness, remitting relenting courses, the presence of speech and hearing problems, more illnesses, restricted activity days, and uncertainty of prognosis could all be considered as risk factors as they alter children's interactions with their environment and the uncertainty may create more strain (Jessop & Stein, 1985; Patterson & Blum, 1996; Nelms, 1989; Ireys, Werthamer-Larsson, Kolodner & Gross, 1994).

The characteristics of illness are often mixed up with severity indicators. Far more correlations are available for measures of these "illness characteristics" or "severity indicators" than for any other measure and more than twice as many correlates of adjustment for illness/disability parameters have been identified than for any other set of variables (Lavigne & Faier-Routman, 1993). Severity is however operationalized in diverse ways and methods differ so that general conclusions are difficult to draw.

All children with a chronic illness and their parents have in common that they are confronted with objective *treatment characteristics*, such as medical visits, therapy or hospitalizations and self-management tasks, although specific features of treatment may not be overlooked. Pantell and Lewis (1987) developed a model to assess the medical impact of medical care on children, in which the medical system influences health through interventions addressing the ability of the child to participate fully in developmentally appropriate activities requiring physical, psychological and social energy.

Not only disease characteristics but also treatment characteristics contribute to the appraisal of the *disease-related events*. Treatment characteristics contribute to changes in the child's and parent's perception and experience of the illness (Maes, Leventhal & De Ridder, 1996) and add to the strain for children as well as parents. Chronic illness and treatment may alter the normal pattern of daily life, for instance because of the necessity to rest, use of medication or having to deal with a diet. The child's

independence can be diminished by the need to have someone looking after the child when swimming or riding a bike or going to school. A time-consuming treatment can increase the risk of developing psychosocial problems, because following the treatment reduces the time available for normal activities, whereas non-adherence can create serious health threats (Patterson & Blum, 1996). A study by Graetz and Shute (1995) revealed children with asthma with frequent hospitalizations were at some risk of peer relationship problems. They concluded that although these children were not actively rejected and able to maintain friendships, their loneliness was worthy of attention.

Demographic characteristics, such as age and gender contribute to the interpretation of illness and the impact on the child and family development. The child's cognitive, emotional and social abilities increase during childhood and adolescence; developmental as well as adaptive tasks change from childhood into adulthood. The impact of illness in childhood is dynamic as it occurs within the context of ongoing developmental processes. For instance, certain age periods constitute a critical period in adherence. Adolescence is, for instance, for patients with diabetes mellitus, a particular difficult time to achieve good metabolic control (Delamater et al., 1991). There is a growing recognition that the children's cognitive-developmental status plays a role in their knowledge, attitudes and behaviors related to health and illness as well as in their emotional response. The age effect on behavior and emotional functioning has however very rarely been studied longitudinally (Thomas & Gustafson, 1996). Longitudinal studies can offer more understanding of the influence of age on stress, coping and outcomes. Gender may also differentially influence outcomes. In general males are reported to have more developmental problems than females (Gortmaker, Walker, Weitzman & Sobol, 1990; Lavigne & Faier-Routman, 1993; Patterson & Blum, 1996). Illnesses that reduce functional physical skills and general strength are reported to be of more concern to males, whereas facial deformity and body size may be of more concern to females (Harper, 1991). The relationship between gender and other variables, such as visibility and disability and behavior and adjustment problems is however complex (Pless & Nolan, 1989).

The appraisal of the diagnosis is also determined by *goals or values* according to expectancy-value theory. Expectancies can be defined as people's degree of confidence of attaining their goals (Maes, Leventhal & de Ridder, 1996, p.232). Problems arise when children want to pursue their goal or to disengage but cannot because of a situational demand. Confrontation with a chronic illness can imply that a child cannot continue to pursue his/her goals (Maes, Leventhal & de Ridder, 1996). The more abilities of high value to a child are threatened by an illness, the more stress the child will experience. Having a chronic illness might imply that plans for future education or career have to be changed. The more goals are threatened and the more important these goals are, the more stressful the experience will be. A child that loves physical exercise and would like to be a sports champion will be more stressed when

confronted with a physically disabling illness, like for instance asthma or juvenile chronic arthritis, than a child who prefers more sedentary activities.

External resources consist of time, money and distance from professional help as well as the social support on which a patient can rely. The family of a child diagnosed with a chronic illness is confronted with additional care-giving demands, the need to integrate the management and care into the family's routines, to help the child in participating in normal activities of childhood, to relate to medical staff and to deal with emotional reactions to the illness, as well as additional financial costs. Family resources of time, energy and money may be directed primarily to the needs of the child with a chronic illness, which may be adaptive for the child and family in the short run, but during an extended period of time other family needs may go unfulfilled (Mac Lean, 1988; Patterson & Blum, 1996).

Families cope with this added strain in a variety of ways and the effectiveness of the family's method of coping will influence the child's coping process as well as its adjustment. Research of Timko, Baumgartner, Moos and Miller (1993) indicated that the strain experienced by mothers predicted in turn the psychosocial and school-related functioning of children. Paternal attitudes and behavior seem to be important as a buffer against maternal dysfunction. Very little, however, is known about how men adjust to child's illness or which coping strategies they employ. It is well documented that they play a lesser role in everyday care and tend to be less informed about the illness (Eiser, 1992). This might explain why Bossert, Holaday, Harkins and Turner-Henson (1990) in a study among children with asthma found that mothers rated their child as more disturbed than fathers or nurses did.

Not only the child and parents, but also the siblings are affected when a chronic illness enters into family life. For the siblings, increased parental and patient dysfunction, more family stressors and less family cohesion and expressiveness are associated with more problems (Daniels, Moos, Billings & Miller, 1987).

Family responses to the chronic illness of the child can either magnify or moderate the risk of psychosocial problems that children with a chronic illness may experience. Good parental functioning, low family stress, a supportive and expressive family environment, family flexibility, adaptive coping, social integration, positive meanings ascribed to the illness, good communication and clear boundaries all appear to be critical variables (Daniels, Moos, Billings & Miller, 1987; Holden, Chmielewski, Nelson & Kager, 1997; Zimmerman, 1995; Patterson & Blum, 1996). Concepts of a family life cycle, which refer to an underlying order of life course which proceeds across phases of development and transition periods, are essential to understand the interaction between illness type, time phase and family functioning (Zimmerman, 1995). There have, however, been no empirical studies reported that have systematically examined these reciprocal relationships between family functioning and the

functioning of children with chronic illness over time (Patterson & Blum, 1996). It is possible that there is a circular feedback loop where the child's positive functioning helps to maintain family life as optimally as possible. Nevertheless, certain characteristics of a parent or family may be risk factors that should alert the clinician to the association between severity and poor adjustment of the child with a chronic illness.



Next to familial support, external resources for children can be found in social support from peers. There are surprisingly few studies on peer relations among children with a chronic illness and their influence on the adaptation of the child. These relations are strained because of the illness, but are important in relation to the stress that children may experience and thus can be a risk or resistance factor in later adjustment (La Greca, 1990b).

Internal resources consist of the energy or physical strength a person possesses as well as personality characteristics such as intelligence, trait anxiety and self-efficacy. These different aspects affect coping behavior. Studies have emphasized that an easy, more sociable temperament is associated with better outcomes. One of the confusing findings is, however, that personality characteristics, such as self-esteem, are, on the one hand, measured as a health outcome, and on the other hand, as a protective factor (Patterson & Blum, 1996).

Actual coping behavior is not only the result of demand or goals appraisals but also depends on demand-resources and goal-resources appraisals. It is important to differentiate between coping actions to deal with the illness and a coping function that refers to the goals these actions intend to achieve. Only a hierarchical model of coping can adequately deal with these different concepts. At the highest level, individuals would then dispose of generalized coping intentions, preferences or dispositions. At the intermediate level, coping strategies occur, whilst the lowest or behavioral level consists of specific coping acts and responses (Maes, Leventhal & de Ridder, 1996). At the higher-order level a distinction is made between problem- and emotion-focused coping. The first type of coping refers to efforts to change or master some aspects of the environment, the other person or the relation between these two elements that is perceived as stressful. The second type of coping refers to efforts to manage or regulate the negative emotions associated with the stressful episode (Compas, Worsham & Ey, 1992). Although this approach may be too general to assess actual illness-specific coping behavior, it facilitates comparison on coping effectiveness (Maes, Leventhal & de Ridder, 1996).

Coping effectiveness implies a relationship between coping behavior and various outcomes. In general, the results of studies on coping among pediatric populations have favored the efficacy of more active, problem-focused efforts over more passive emotion-focused coping (Compas, Worsham & Ey, 1992). With regard to the

development of coping skills during childhood and adolescence, studies suggest that problem-focused coping and emotion-focused coping skills emerge at different points in a child's development. Problem-focused skills appear to be acquired earlier, with some evidence for the acquisition of problem-solving skills apparent in preschool years. One reason for this may be that they are more readily acquired through modeling of adult behavior as many of the involved coping strategies imply overt behavior. Emotion-focused coping skills appear to develop in later childhood and early adolescence when there is a more advanced cognitive development (Compas, Worsham & Ey, 1992). It is, however, premature to conclude that emotion-focused coping is generally an ineffective strategy (Compas, Worsham & Ey, 1992). Further examination of the role of coping in affecting the child's adjustment is recommended (Lavigne & Faier-Routman, 1993; Röder, in preparation).

For children with a chronic illness, physical symptoms or complaints are present, treatment is life-long, and they are confronted with (daily) self-management tasks and limitations in daily life. The *physical consequences* for children with a chronic illness thus relates to 1) the physical symptoms or complaints, 2) the ability to participate in physical activities as well as 3) the aspects of (daily) treatment that is oriented towards amelioration or maintaining stability of the physical illness.

Illness-specific, as well as general symptoms or complaints, belong to indicators of the physical illness. General somatic complaints are more widely reported among children with a chronic illness as compared to a healthy group (Wallander, Varni, Babani & Banis, 1988). Illness symptoms have been often used as a severity measure to predict outcomes. It is recommended that severity should be used as a measure of medical status within a diagnostic group and presumed to reflect the child's underlying physical illness.

Depending on the characteristics of the illness, the severity of symptoms, the treatment available and other mediating variables, children are handicapped in doing their regular daily activities such as going to school or participating in outdoor activities. Functional status is often defined as the degree to which the child can perform daily tasks at an age-appropriate level. As school is the place to receive education and herewith prepare for a job as well as the place to meet other children, going to school is an important protective factor.

The current trend is to give the patients (or their parents) an increasing role in the management of their chronic illness. Children from 8 years and older start to learn their self-management tasks. These self-management tasks may be necessary for life time, as in diabetes where children have to learn to control their blood glucose level through regular controls and diet interventions. Adherence, capabilities and emotions concerning self-management are all important psychological issues. Pediatricians,

nurses, psychologists have to work in a team to improve this part of the care for children with a chronic illness.

Chronic illness can influence *social functioning* either as a direct consequence of illness symptoms or indirectly through limitations imposed by parents. According to La Greca (1990b), pediatric illnesses in which the social consequences are most pronounced are illnesses that imply: (1) a restriction of physical activities, (2) interruptions of daily activities, (3) altered physical appearance, either by illness or treatment, or (4) modifications of lifestyle, a strict program of exercise, diet or taking medication (La Greca, 1990b).

Given the limitations of some of the children with a chronic illness, it is understandable that they have more difficulties in participating in social activities than healthy children or that they have fewer contacts with friends (Wallander, Varni, Babani & Banis, 1988; Cadman, Boyle, Szatmari & Offord, 1987). The child's illness may further impact on peer relationship development through its impact on heightened parental restriction of peer activities (Nassau & Drotar, 1995).

The elementary school provides opportunities for peer interactions and treatment and illness may form barriers to these. Children with a chronic illness may report negative perceptions about school primarily resulting from peer teasing and social isolation. In a study among mothers of children with a chronic illness, 35,4% reported discrimination of which more than half of them associated with school. Furthermore 15% of the mothers experienced difficulty in taking children to public places because of structural, legal and attitudinal barriers (Turner-Henson, Holaday, Corser, Ogletree & Swan, 1994). In a study among school-age children, 14% of the parents of children with a chronic illness indicated that their child was confronted with peer problems such as teasing, isolation or even hitting. In addition, parents of other children may not allow the child to sleep overnight because of the increased responsibility (Turner-Henson, Holaday, Corser, Ogletree & Swan, 1994). Peer difficulties may start early in life. A study among preschoolers revealed that preschoolers preferred a healthy child above a child in a wheelchair when they had to choose someone to play with in active games (Nabors & Keyes, 1997).

Conformity becomes important in the primary school group and removal from peers is therefore a risk factor (Spirito, DeLawyer & Stark, 1991; Graetz & Shute, 1995). It is suggested that placement in a normal school can be most conducive to achieving good academic standards, but that children might be happier in situations where they are in the company of other children suffering from similar illnesses (Lord, Varzos, Behrman & Wicks, 1990).

Youngsters with a chronic illness are likely to encounter social situations that differ from those of their healthy peers. Compared to children who are healthy, children

with a chronic illness are more frequently in contact with adults other than parents or teachers, because of medical examinations or hospitalizations. Whether this will also have beneficial effects on children is not clear.

A review of literature on social adjustment revealed inconclusive results on the quality of peer relations among children with a chronic illness. Empirical work has been limited and such research as is available suffers from inadequate methodology. Due to contradictory findings it is unclear whether differences exist between chronically ill and healthy children in the quality of peer relations (Spirito, DeLawyer & Stark, 1991). Peer relations in childhood are however important as they are predictors of adjustment, future competence during adolescence and adaptation into adulthood (Turner-Henson, Holaday, Corser, Ogletree & Swan, 1994). In addition, the interference of illness in a child's social life is an important factor for non-adherence, of which parents are often not aware (La Greca, 1990b). The assessment of social consequences seems therefore important to identify children at risk.

Psychological consequences are defined by cognitive and emotional or behavioral consequences. Children spend a lot of time on academic tasks. Academic schooling prepares the child for job-related tasks in the future. While there is an indication that children with a chronic illness are at some risk academically, there is actually no simple explanation for this. School absence, illness characteristics (e.g. neurological impairments) and side effects of medication may have a direct influence on cognitive functioning, whereas lower expectations from parents or teachers, overrestrictive parenting, learned helplessness and social isolation may influence the academic achievement indirectly.

School absence appears to be higher for children with a chronic illness compared to healthy children (Zimmerman, 1995). It has been reported that mean days of absence for children with chronic illness (Johnson & Atkinson, 1985) were 16 days compared to less than 7 for healthy children (Eiser, 1992).

Lower expectations caused by children's dependency or the overprotectiveness and pessimism of both parents and teachers have also been reported to contribute to educational underachievement (Kim, 1991). On the other hand, Peckham, Meadows and Marrerro (1988) suggested that the wide range of achievements shown by children with a chronic illness, indicate that supportive environments can do much to overcome disadvantage.

Behavioral and to a greater extent, emotional consequences have often been studied in relation to adaptation to illness and are considered to be highly relevant as outcome measures. It is believed that psychosocial problems occur twice as often among children with a chronic illness than among healthy children (Wallander, Varni, Babani & Banis, 1988; Eiser, 1990b; Barbarin, 1990; Austin, 1989; Perrin, Ayoub & Willett, 1993; Ireys, 1994). The large and often cited epidemiological Ontario Health Study (OHS) in

the general community in the Canada revealed that children with both chronic illness and associated disability were at a more than triple risk of psychiatric disorders. Children with medical illness but no disability were at less risk: about a twofold increase in psychiatric disorders (Cadman, Boyle, Szatmari, & Offord, 1987).

The understanding of adaptation problems is however difficult as adaptation has been conceptualized and operationalized in numerous ways, ranging from self-esteem to specific symptoms and problems (e.g., anxiety, depression, activity level) (Thompson & Gustafson, 1996). Most studies look for correlations among variables to understand why children are adapting less well than comparison groups. Positive effects are however hardly ever studied (Barbarin, 1990). In addition, one complicating factor is that some variables are considered to be independent as well as dependent variables, like self-esteem, so that studies are difficult to compare (Lavigne & Faier-Routman, 1993). Differences among different diagnostic groups are not clear. Internalising problems have been most often studied among children with a chronic illness, and appear to predominate in this group (Harris, Canning & Kelleher, 1996).

1.8 CONCLUSION

Given the number of children with a chronic illness, the limitations that children have to cope with, and the variety of consequences, it is not surprising to find health psychologists increasingly involved with research, education and clinical practice in pediatrics. Although very common in adults, research on stress, coping and quality of life using stress-coping models as a theoretical framework among children with a chronic illness has been started only recently. Research guided by these models may however lead to more knowledge about risk and resistance factors, which may offer the clinical practitioners useful tools for developing successful prevention and intervention programs (Lemanek, 1994).

In this chapter the stress and coping model developed by Maes, Leventhal and DeRidder (1996) was used as a framework to review variables thought to influence the consequences of a childhood chronic illness. Although this model was developed for studying stress and coping among adults, this model was chosen for several reasons. First of all, it was felt that the variables used for adults might in general also apply to children, although they have to be operationalized differently. Furthermore, it was felt that the clear description of the variables that are important in coping with a chronic illness as well as the extension with variables such as other life events and life goals or values made it a useful theoretical framework for describing determinants of coping with childhood chronic illness. For instance, other life events, such as divorce of parents or change of school, can influence the appraisal of a chronic illness, increase the stress children experience and increase the risk of difficulties in functioning. Goals or values held by the individual child can influence the interpretation of illness. The

more abilities of high value are threatened, the more stress a child will experience and the more the child will be at risk. It must be borne in mind, however, that the process of stress and coping is not static but dynamic especially in children as their age and the family life cycle are progressing rapidly. These aspects are not described in the model and have to be taken into account when studying the process of adaptation to childhood chronic illness.

A delineation of the consequences on physical, social and psychological functioning was given. The main aspects of physical consequences are general physical symptoms or complaints, the ability to participate in physical activities and aspects of daily management tasks. Social consequences refer to the ability to participate in social activities but also to the social problems children may experience, such as being teased or left alone. Psychological consequences refer to the cognitive, emotional and behavioral problems children may experience. Cognitive consequences refer to the ability to fulfill age-related cognitive tasks and the ability to participate in school lessons. As emotional consequences rather than behavioral consequences seem to be of greatest importance for children with a chronic illness, the main focus will be on emotional consequences in this doctoral dissertation.

Depending on risk and resistance factors, the child's functioning might be influenced more or less on all these dimensions. Knowledge about the consequences of childhood chronic illness is helpful when diagnosing children in need of intervention and when evaluating interventions.

CHAPTER 2: CONSEQUENCES OF CHILDHOOD CHRONIC ILLNESS

2.1 INTRODUCTION

In this chapter a review of major findings in clinical studies focusing on the physical, social and psychological consequences for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis are described. To facilitate the understanding of the adaptive-illness-related tasks with which the child and family are confronted, symptoms and treatment of each of the selected illnesses is first described.

2.2 CHILDHOOD ASTHMA

2.2.1 Definition, prevalence and characteristics

Asthma is difficult to define because no single definition has been acceptable to everyone (Thompson & Gustafson, 1996, Evans, 1994). The Dutch "NHPG guidelines for GPs 1992" define asthma as follows: "Asthma can be diagnosed in children when at least one of the following symptoms is relevant: periods of coughing and/or congestion of the chest that occur at least five times a year and when not adequately treated last at least ten days, attacks of breathlessness and mainly wheezing, shortness of breath or wheezing after physical exercise". Until more information is available, the definition of asthma must be a clinical one based on recurrent wheezing (Price, 1994).

As described in the first chapter, asthma is the most common childhood chronic illness affecting 9% of children aged 0-14 years in the Netherlands. Boys are more affected by asthma than girls (Wulp, 1996). As many as 75% of the children have a family history of asthma. Asthma in children usually begins between 3 and 8 years of age and symptoms are usually caused by an allergy. The child may also have other symptoms as eczema.

Asthma is characterized by airway obstruction resulting from muscle spasm in the lungs, inflammation and swelling of the bronchial tubes or excess mucus. It is generally accepted that the underlying defect is hyperreactive airways and that allergy play a role in this reaction (Spykerboer, Donnelly & Thong, 1986). The acute airway obstruction remits either spontaneously or with treatment (Creer & Bender, 1995). Although 60% of children younger than one year have episodes of coughing and wheezing, fewer than half of them develop asthma at six years. Eczema, and family history for atopy heightens the risk of having asthma at six years (Hoekstra, 1997). Whilst a long-term prognosis is difficult, children with asthma have a greater risk of developing chronic aspecific respiratory diseases at an older age, although this risk can be decreased by the right treatment (Brandt & Monnens, 1990).

The number of attacks varies both from patient to patient and from time to time. Most children have episodes with asthma with asymptomatic periods between episodes (Richards & Hemstreet, 1994). The symptoms can vary with the season of the year and across lifespan. Asthma attacks often occur at night and may be precipitated by exposure to allergens during the day. Physical exercise, stress and emotions can also induce shortness of breath. An acute, more severe and long-lasting attack is called status asthmaticus. This can cause a lack of oxygen in the blood, blue skin, and unconsciousness, which is a medical emergency (Thompson & Gustafson, 1996). Variability refers to the overall severity of a patient's asthma as well as to the intensity of discrete attacks (Creer & Bender, 1995).

2.2.2 Treatment

Although asthma would appear to be a condition that is, in many cases, treatable in general practice, studies have shown that many of the children are underdiagnosed or late diagnosed and untreated in this setting (Hilton, 1994). The treatment of asthma includes both the prevention of attacks and the management of attacks. In order to prevent attacks, environmental control methods are often recommended. These include decreasing exposure to dust and dust mites, and to pets and cigarette smoke. In order to manage attacks, pharmacological treatment is used involving three kinds of drugs: bronchodilators, which relax and open the airways; anti-inflammatory agents, which decrease swelling and allergic reactions in the lungs; mast-cell stabilizers, which inhibit the production and release of substances that create allergic reactions in the lungs (Thompson & Gustafson, 1996). Although reports of psychological side effects of medications are prone to exaggeration, some do exist and need to be considered when conducting research or planning interventions among children with asthma (Creer & Bender, 1995). Effective management depends on medication and self-management capabilities (Thompson & Gustafson, 1996). Comprehensive management of a chronic illness in children requires both patient and family involvement (Moe, Eisenberg, Vollmer, Wall, Stevens & Hollis, 1992).

2.3 INSULIN DEPENDENT DIABETES MELLITUS

2.3.1 Definition, prevalence and characteristics

Insulin dependent diabetes mellitus (IDDM) was first described by Demetrius of Apameia in the third century before Christ, who called the disease "diabetes" which means pipe or sifon. In 1674 an English physician Thomas Willis, discovered the sweet taste of the urine and it was probably then that the word "mellitus" (sweet flow) was added (Peltenburg, 1995).

Diabetes mellitus most often develops in childhood and affects 0.2% of the children in the Netherlands (CBS). It is estimated that there are around 337 new cases under 15 years of age per year or 12.4 per 100.000 (Ballegooie, 1995). Compared to other

European countries this is a low prevalence, although there has been an increase of new cases of 13% in 5-19 years olds in the ten-year period from 1978-1980 to 1988-1990. In children from 5-9 years, the incidence is the highest among girls in contrast to the other age groups (Ballegoie, 1995). The diagnosis of diabetes mellitus is made most frequently in two age periods: 5-6 years and 11-13 years (Johnson, 1995).

Diabetes mellitus is a complex disorder that is mainly caused by the failure of the pancreas to release enough insulin into the body so that children become dependent on insulin injections. Insulin serves to lower blood glucose levels in the blood by making cell membranes permeable to glucose and allowing it to enter the cells of liver, muscles and fat cells. The beta cells continuously deliver a small amount of insulin and provide an additional quantity after rises in blood glucose levels after a meal. As a consequence of the high blood glucose level, frequent urination occurs so that thirst, the need to drink a lot and loss of weight are the onset signs of diabetes mellitus. It is believed that both hereditary and exogenous determinants together can cause diabetes mellitus (Bruin, 1994).

Despite modern treatment with insulin and the use of simple blood sugar measurement tests, which may help to maintain an optimal blood sugar level, diabetes mellitus is a progressive disease. Primary complications of diabetes mellitus are a high glucose level (hyperglycemia) or a low blood sugar level, called "hypoglycemia". A "hyper" causes extreme thirst, a dull headache, nausea, and fatigue, pain below the breastbone, dry lips, sunken eyes and a fruity breath odor. Sometimes the symptoms are so severe that coma can occur. A "hypo" occurs more often and is characterized by nervousness, sweating, headache and slurred speech. In severe cases unconsciousness can occur. In that case an injection with glucagon is needed (Peltenburg, 1995). Secondary complications can be retinopathy, nephropathy or neuropathy. In addition, diabetes mellitus is a risk factor for heart disease and vascular disease. These complications are related to a high blood glucose level for many years and probably also other factors as hereditary factors (Krans, 1995) and may lead to high illness absenteeism and early death. Life expectancy is still one third less than that of the general population (Johnson, 1988).

2.3.2 Treatment

The goal of treatment in diabetes mellitus is to keep blood sugar levels as close to normal as possible through the administration of insulin injections. Diet, insulin doses and exercise all have to be taken into account to maintain a correct sugar and insulin balance. The interactions of diet, exercise, illness, emotional state and insulin makes the management of diabetes mellitus difficult (Thompson & Gustafson, 1996).

Simple blood glucose tests are available that allow the patient to self-test the blood glucose level. Most children use an insulin pen with which it is possible to inject a dose of insulin several times a day. Insulin is available in two sorts, one with a short-term

effect, the other with a long-term effect. More precise regulation of insulin has been possible since short-term insulin became available.

Where sugar and candies were once forbidden, nowadays the content of the child's diet can be very similar to that of any other child although the intake of saturated fat has to be limited. A child however must always take account of the balance between its blood sugar level, diet (the amount of carbohydrates) and exercise. A diabetic child has to take snacks during the day, which is often seen as an advantage but may also place the child in a special position in class as eating in class is often forbidden. The less strict rules that apply to diet today may also complicate the diet because it is not so clear to the child and other persons what is allowed or not. Adherence can be a major issue because of the daily tasks that have to be accomplished and the difficulty of maintaining metabolic control. In particular, adolescence is often a difficult period (Thompson & Gustafson, 1996).

2.4 EPILEPSY

2.4.1 Definition, prevalence and characteristics

The word "epilepsy" is of Greek origin and means "to seize upon" or to attack (Neppe, 1985; Temkin, 1994). Seizures and epilepsy are clinical phenomena resulting from abnormal and excessive excitability of the neurons of the cerebral hemispheres. An epileptic seizure is an intermittent, paroxysmal, stereotyped disturbance of consciousness, behavior, emotion, motor function, perception or sensation (which may occur singly or in combination) which on clinical grounds, results from a cortical neuronal discharge (Appleton & Gibbs, 1998). Epilepsy is a condition in which seizures recur, usually spontaneously. The clinical event is accompanied by characteristic electro-encephalographic (EEG) changes, the epileptic discharge, which may be local (focal) or diffuse (generalized). It must be accompanied by a clinical change. Thus epilepsy is a clinical and not an EEG diagnosis.

Seizures represent one of the most common types of neurological conditions. Estimates from Rochester, Minnesota, USA, indicate that, by the time people reach their seventies, the risk of having had at least one seizure is about 8% (Hauser & Kurland, 1975). More than fifty percent of the individuals who have seizures have the first (and sometimes only) seizure during childhood or adolescence. Thus, the initial occurrence of epilepsy is concentrated during childhood years. The annual incidence of newly diagnosed epilepsy in children under 20 years is about 0.6/1000. The prevalence of having had a seizure within the past three years or currently taking medication among children is about 4/1000 (Berg, 1995).

There are three axes along which seizures may be classified (Berg, 1995):

1. Provoked versus unprovoked: acute symptomatic (or provoked) seizures occur in the immediate context of a neurological insult (e.g. head injury, encephalitis, hypoglycemia, fever). Unprovoked seizures occur when no such provocation is present: "spontaneously" occurring seizures.
2. Presence of neurological abnormalities (remote or symptomatic) versus idiopathic: children who have an identifiable brain abnormality (e.g. history of a stroke, cerebral palsy, mental retardation) are classified as having symptomatic seizures. "Idiopathic" implies that no structural brain abnormality exists or can be identified.
3. Generalized versus partial onset: seizures which are generalized from onset are referred to as generalized seizures, whereas seizures that begin in a localized region of the cortex (whether or not they spread to involve other areas or even when the whole cortex is involved in secondarily generalized seizures) are called partial or focal onset seizures (Commission of classification and terminology of the ILAE, 1981).

The EEG features are included in this classification, since it is sometimes impossible to classify a seizure as focal or generalized without EEG confirmation. An EEG should be an integral part of seizure evaluation.

All seizures can be classified against these three axes. This will not result in all patients in the diagnosis epilepsy. Epilepsy is characterized by recurrent unprovoked seizures. Thus, an individual with acute symptomatic seizures or a single unprovoked seizure is not considered to have epilepsy. The designation of remote symptomatic versus idiopathic is not a factor in assigning the diagnosis of epilepsy, but is of importance with respect to prognosis.

Generalized seizures are assumed to involve the entire brain simultaneously at the onset of the seizure. Most common are the tonic-clonic seizures and absences. Tonic-clonic seizures (old terminology: grand mal) present without warning with a loss of consciousness and the tonic component, consisting of symmetric fixed extensions or flexion posturing of the limbs. The child may clench the teeth, bite the tongue and lose bladder control. During this stage the child does not breathe and may emit a cry. This is followed by rhythmic jerking of the extremities (clonic phase). The tonic-clonic sequence is variable. At the end of the seizure the jerks fade away, consciousness returns and the child often falls in a deep sleep. Some children have a headache and feel tired; others are remarkably alert within a few minutes.

Typical absence seizures ("petit mal") are described as brief periods of loss of awareness with staring occasionally accompanied by a few eyelid twitches or other little movements (e.g. motor automatisms of the hands). Onset is without warning and children have a complete amnesia for what has happened and after the seizure

continue with what they were doing (Ansink, 1980; Sachs, 1995). Absences practically occur only in children. A certain percentage of children with absences also experience tonic-clonic seizures.

Within the category of generalized seizures, there are a number of subgroups including myoclonic seizures. Myoclonic jerks, sudden brief contractions of single muscle groups which may be repetitive, are the clinical representation of myoclonic seizures (Sachs, 1995).

Partial seizures are further subdivided into those that do not impair consciousness (simple partial seizures) and those in which consciousness or alertness is impaired (complex partial seizures). Simple partial seizures may present with motor, sensory, autonomic or rarely psychic symptoms. Complex partial seizures may begin as simple partial seizures or as complex seizures from the outset, and show motor phenomena as walking around or other purposeless, involuntary automatisations such as picking at the clothes. All partial seizures may evolve into (secondarily) tonic-clonic seizures.

2.4.2 Treatment

In the past decades the way physicians think about seizures has changed. The possible risks of treatment (potential adverse effects of medication) and the fact that even prolonged seizures rarely cause brain damage has led to the insight that not all children with seizures of any type have to be routinely treated with anti-epileptic drug (AED) therapy. In addition, children with newly diagnosed epilepsy achieve in approximately 70-75% remission (a period of time - 6,12 or 24 months - without seizures), some of them even without AED therapy. Informed decision-making regarding initiating (or withdrawing) AED therapy must consider both the (statistical) risk of further seizures and the potential adverse consequences of seizure recurrence, versus the possible adverse effects of AED therapy.

The current AEDs have proven to be effective in the treatment of the majority of children, but are not without side effects. Detrimental effects on cognitive functioning, activity level, behavior control and mood have been noted. AEDs, the epilepsy itself and the psychosocial situation may all contribute to these problems. This requires close medical monitoring and intensive communication between the child, the parents, the physician and the school teacher (Sachs, 1995).

The goal in contemporary epilepsy treatment is to achieve complete control of seizure activity without side effects with monotherapy. Treatment with medication cannot offer a cure but is aimed at prevention of seizures so that the patient can lead a nearly normal life. In up to 70-80% of children, attempts are generally made to stop medication after one or two years without seizures. After withdrawal of medication, 60 to 75% of patients remain seizure free (Rutgers, 1989; Peters, Brouwer, Geerts, Stroink, Donselaar, 1998). Relatively few restrictions are necessary for children with epilepsy

(O'Donohoe, 1983). However, certain activities do require specific cautionary comments, such as swimming, bathing, cycling, climbing. It is of great importance that physicians reduce restrictive advises as soon as the seizures are completely controlled.

2.5 JUVENILE CHRONIC ARTHRITIS

2.5.1 Definition, prevalence and characteristics

Rheumatic symptoms such as limping and limb pain, are common in children, but chronic arthritis in children is rare. Juvenile chronic arthritis (JCA) is a condition thought to be caused by abnormal immunological mechanisms affecting the connective tissues particularly those of the joints. About 1 child in every 1000 has juvenile chronic arthritis (Soesbergen, 1991; Southwood, 1995). Each year one new case will be expected in every 10.000 children (Southwood, 1995). In the Netherlands it is estimated that 500-2000 children have juvenile chronic arthritis (Soesbergen, 1991). Juvenile chronic arthritis is the most prevalent of the rheumatoid diseases in childhood. To diagnose juvenile chronic arthritis, the onset must be before the age of 16, the arthritis must be active for more than three months and other disease must be excluded (Southwood, 1995). Chronic inflammation and swelling of joints are associated with stiffness, pain and may cause permanent damage. Symptoms vary from day to day and from morning till afternoon.

The diagnosis is difficult because swelling of joints may be subtle or absent initially and complaints of pain may be difficult to obtain from children. Children may present with limping, upper limb dysfunction, torticollis or non-specific constitutional symptoms such as lethargy, fever or poor appetite. Occasionally features not associated with the musculoskeletal system may predominate. Prompt diagnosis is however important to prevent joint contractures, muscle wasting and abnormal growth (Southwood, 1995).

The basis for the classification of juvenile chronic arthritis is the number of joints affected during the first six months of the disease and extra-articular clinical features. Juvenile chronic arthritis may best be conceptualized as a syndrome with 3 distinct subtypes of disease onset (Varni, Thompson & Hanson, 1987).

The pauci-articular onset; the most common subtype of juvenile chronic arthritis, accounts for over half of the cases and usually affects young (preschool) girls. It is characterized by arthritis in four or fewer joints (Varni, Thompson & Hanson, 1987). It is commonly associated with antinuclear antibodies. The prognosis of the patients is good for most of the children. In most cases the arthritis is not severe and only persists for a few years. One third of the children, however, will develop polyarthritis that is more difficult to control. Two other groups may also present with pauci-articular disease; those with juvenile psoriatic arthritis (skin disease with inflammation of the

joints) and those who are predisposed to ankylosing spondylitis (stiffness of the spinal column) (Soesbergen, 1991; Southwood, 1995).

The polyarticular onset is predominantly symmetrical and affects upper and lower limbs and is found in 30-40% of the children with juvenile chronic arthritis. This form is characterized by arthritis in five or more joints. Most patients are girls who develop arthritis during their preschool years. In less than 10% of the cases of juvenile chronic arthritis, polyarticular arthritis starts in late childhood and adolescence. Both groups have a poorer prognosis than those with pauci-articular onset do (Southwood, 1995).

The systemic onset is the rarest form of juvenile chronic arthritis. Most patients develop disease in early childhood but patients of any age may be affected. It is often difficult to diagnose. This form is characterized by intermittent fever with or without rheumatoid rash or other organ involvement (Varni, Thompson & Hanson, 1987). The number of arthritic joints varies: about a third of patients develop severe polyarthritis that is resistant to treatment and the articular outlook for these patients is poor (Southwood, 1995).

2.5.2 Treatment

Early diagnosis and treatment improves prognosis (Southwood, 1995). Affected children rarely need hospitalization but hospitalization is indicated when the child is unable to walk, has progressive flexion deformities unresponsive to physiotherapy or severe extra articular features of disease. Progress needs to be monitored carefully. Non-steroidal anti-inflammatory drugs (NSAIDS) are indicated in children with active synovitis. Side effects of these drugs include abdominal pain, changes in mood and rash. Intra-articular corticosteroids are indicated for arthritis that is not controlled by non-steroidal anti-inflammatory drugs (Southwood, 1995). Treatment also includes, in general, a balanced program of exercise and rest. Children may need splints to prevent contortion of joints or to rest the joints. Tools like a special pen can be beneficial when joint limitations are apparent. It is sometimes necessary to perform a synovectomy (a cleaning operation) or replacement of joints, although this is fortunately not often needed in children (Soesbergen, 1991).

2.6 CONSEQUENCES OF ILLNESS ON THE CHILD'S FUNCTIONING, THE REVIEW METHOD

In the following paragraphs, main findings of clinical studies on the physical, social and psychological functioning of children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis are reviewed. Studies were searched via the PsycLit and Medline CD-Rom system. This search was conducted with the search terms: child*, physical, cognitive, social, emotional, years 1987 to June 1997 and revealed 564 articles on children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. Articles in the personal file of the author were added. Subsequently, the

articles were selected according to the following selection criteria: (a) the articles had to include clinical studies on children including primary school children, so that reviews or papers on instrument development or intervention evaluation were excluded, as were case studies; (b) the physical, social and psychological functioning of the child as defined in chapter one had to be the main focus of the article; (c) the children included in the population studied had no other major handicaps such as mental retardation or psychiatric disturbances; (d) only papers in German, English or Dutch were included. In addition, studies using only associative tests like Rorschach or Draw a Person Test were excluded.

In this review the types of problems within the defined dimensions in each of the four selected groups and the general results on these dimensions of functioning are described. In paragraph 2.7 to 2.10 clinical studies comparing the four groups with healthy children or population studies are described. In paragraph 2.11 a description of studies comparing children with different chronic illnesses is given.

2.7 THE FUNCTIONING OF CHILDREN WITH ASTHMA COMPARED TO HEALTHY CHILDREN

Following the selection criteria described above, 20 articles on children with asthma were selected (Bremberg & Kjellman, 1985; Viney & Westbrook, 1985; Suess, Stump, Chai & Kalisker, 1986; Marx & Florin, 1986; Chiari, Foschino, Nuzzo, Pecci & Rossi, 1987; Eiser & Town, 1988; Perrin, MacLean, Gortmaker & Asher 1989; Furrow, Hambley & Brazil, 1989; Hambley, Brazil Furrow & Chua, 1989; Gutstadt, Gillette, Mrazek, Fukuhara, LaBrecque & Strunk, 1989; Weston, MacFarlane & Hopkins, 1989; Townsend, Feeny, Guyatt, Furlong, Seip, & Dolovich, 1991; Groot, Boer & Baecke, 1992; MacLean, Perrin, Gortmaker, Pierre 1992a; Butz & Alexander, 1993; McNaughton, Smith, Rea, Asher, Mitschell, Mulder, Seelye, Stewart & Taylor, 1993; Meijer, Griffioen, Nierop & Oppenheimer, 1995; Graetz & Shute, 1995; Eksi, Molzan, Savasir & Guler, 1995; Bussing, Burket & Kelleher, 1996). Most studies included the *physical or emotional functioning of children with asthma*. Nearly half of the studies included cognitive functioning and only four studies were directed to the social functioning of children with asthma (see Table 2.1 in Appendix 1).

With regard to *physical functioning*, children with asthma are reported to have relatively more physical symptoms or complaints compared to healthy children (MacLean, 1992a, Furrow, Hambley & Brazil, 1989). For instance, 32% of the children with asthma in a study by Townsend, Feeny, Guyatt, Furlong, Seip, & Dolovich (1991) cited difficulty in falling asleep, not being able to sit still and restlessness as a consequence of illness or medication. In addition, children with moderate asthma found their asthma symptoms troublesome, respiratory symptoms were cited frequently as most bothersome. Children with asthma are reported to have more limitations in physical activities than healthy children (Bremberg & Kjellman, 1985;

Townsend, Feeny, Guyatt, Furlong, Seip, & Dolovich, 1991). For instance, it was found that 78% of the children were reported by their parents as being restricted in physical education at school (Bremberg & Kjellman, 1985). For physical activities, running was reported to be the most frequently affected activity and 85% of the children listed this as problematic (Townsend, Feeny, Guyatt, Furlong, Seip, & Dolovich, 1991), though contrasting findings were found by Hazzard and Anger (1986) and Weston, MacFarlane and Hopkins (1989). The study by Hazzard and Anger (1986) using parent reports, revealed that children with asthma were fairly active, exercising at least several times a week. These results were confirmed by Weston, MacFarlane and Hopkins (1989), who found in child reports on physical activities that children with asthma were more frequently active and more anxious prior to exercise than healthy children but had no higher perceived level of activity. The children with a higher prevalence of asthma enjoyed a lower level of fitness and felt asthma deterred them from doing more exercise, but had an equal prevalence of activities (Weston, MacFarlane & Hopkins, 1989). A possible explanation could be that, although children with asthma may participate as much as healthy children in physical activities, they may be hindered in their performance on these activities.

Three studies discussed daily management tasks (Hazzard & Angert, 1986; Eiser & Town, 1988; Mc Naughton, Smith, Rea, Asher, Mitschell, Mulder, Seelye, Stewart & Taylor, 1993) indicating that effective management was facilitated in conditions with low stress, social support and little need of contact with official agencies. Although Eiser and Town (1988) found no age effects on knowledge and control of health status, the study by Hazzard and Angert (1986) revealed that increasing age and education made children more knowledgeable and more capable of controlling their health status. How children experience these daily management tasks was not examined in the selected studies.

In respect of *social functioning*, children with asthma had a lower social competence score than healthy children on the Child Behavior Checklist (CBCL) (Eksi, Molzan, Savasir & Guler, 1995). Inspection of overall CBCL social competence scale scores revealed that 5% of the children had scores regarded as clinically significant. In addition, a higher severity led to more social problems (MacLean, Perrin, Gortmaker, Pierre 1992a). On the contrary, Graetz and Shute (1995) found no differences in social competence or popularity, reciprocated friendships or self-perceived loneliness between children with asthma and healthy children, although frequent hospitalizations were reported as a possible negative factor in this.

As the CBCL mainly measures participation in social activities rather than the competence of children to establish and maintain friendships, a possible explanation for these contrasting findings is that children with asthma do experience limitations in social activities but that, in general, they experience no more problems than healthy children in social relations.

With regard to *cognitive functioning*, most of the selected studies report limitations at school for children with asthma. Absenteeism is reported to be higher for children with asthma as one-third of all school absences is related to airway problems (Groot, Boer & Baecke, 1992), which might have an impact on school functioning of the individual child. School attendance may further be influenced by other factors than asthma alone, such as school, economic and cultural variables. Performance scores decreased when lower socioeconomic status, older age, continuous oral steroid use, higher severity and when emotional or behavioral problems were present (Gutstadt, Gillette, Mrazek, Fukuhara, LaBrecque & Strunk, 1989; Mc Naughton, Smith, Rea, Asher, Mitschell, Mulder, Seelye, Stewart & Taylor, 1993). According to a study by Bremberg and Kjellman (1985), 11% of the children with asthma experience restriction in cognitive functioning during normal lessons at school.

Most children with asthma, especially younger boys and adolescents, were experiencing school-related problems as measured with the CBCL in a study by Hambley, Brazil Furrow and Chua (1989). Bussing, Burket and Kelleher (1996) indicated that children with asthma had more past school problems, school refusal and repeated grades or special school placements than healthy children. A lower performance on school competence as measured by the CBCL was also found for children with asthma compared to healthy children (Eksi, Molzan, Savasir & Guler, 1995).

Medication can be a factor in this, as steroid theophylline medication can affect cognitive performance for 6 hours after intake (Suess, Stump, Chai & Kalisker, 1986). In addition, one-third of the children cited difficulty in falling asleep, not being able to sit still and restlessness as a consequence of illness or medication (Townsend, Feeny, Guyatt, Furlong, Seip, & Dolovich, 1991) which may also have an impact on school functioning.

In contrast, Gutstadt, Gillette, Mrazek, Fukuhara, LaBrecque and Strunk (1989) found that children with asthma had average or above average academic abilities, even though children were absent from school one fifth of the days prior to testing. Although school absence is relatively high and children may experience difficulties in school functioning as a (indirect) consequence of their illness symptoms or medication, there is no agreement on the cognitive consequences children with asthma may experience.

A greater number of *internalizing problems* were found in children with asthma compared to healthy children (Furrow, Hambley & Brazil, 1989). MacLean, Perrin, Gortmaker, Pierre (1992a) found a higher score on internalizing than on externalizing behavior problems in children with asthma, while clinical elevations were higher for internalizing than for externalizing problems. According to Bussing, Burket and Kelleher (1996) children with asthma experienced more anxiety, more

psychiatric illnesses and more interfamilial stress than healthy controls. Conclusions on severity are not easy to formulate as children with mild asthma and severe asthma had significantly lower adjustment scores than children with moderate asthma in a study by Perrin, MacLean, Gortmaker and Asher (1989), and in a study by Eksi, Molzan, Savasir and Guler (1995) severity was not a mediating variable. Gender may play a role as girls had a lower self-esteem than boys (Hambley, 1989).

There were however also studies that found no negative emotional or behavioral consequences for children with asthma or even positive consequences. In contradiction to what was presumed previously, Viney and Westbrook (1985) found no suppressed emotional reactions among children with asthma, Marx and Florin (1986) too did not find a deficit in facial reactions. Families exhibited normal levels of bonding and flexibility in response to stress (Hambley, Brazil Furrow & Chua, 1989). As a group, children with asthma had normal emotional adjustment scores (Perrin, MacLean, Gortmaker & Asher, 1989). A higher internal locus of control and a higher self concept was found among children with asthma than healthy children (Hazzard & Angert, 1986).

In conclusion, children with asthma are reported to have more physical complaints and more physical limitations than healthy children. In addition, they experience more limitations in social activities than healthy children. Although the school absence is relatively high for children with asthma and school difficulties have been reported, there is no agreement on the consequences for cognitive functioning that children might experience. There are no conclusive results found for the emotional functioning of children with asthma.

2.8 THE FUNCTIONING OF CHILDREN WITH DIABETES MELLITUS COMPARED TO HEALTHY CHILDREN

Following the selection criteria described above, 35 articles on children with diabetes mellitus were selected (Holmes & Richman, 1985; Gross, Delcher, Snitzer, Bianchi, & Epstein, 1985; Ryan, Longstreet & Morrow, 1985; Kovacs, Brent, Steinberg, Paulauskas, & Reid, 1986; Johnson, Silverstein, Rosenbloom, Carter & Cunningham, 1986; Rovet, Ehrlich & Hoppe, 1987; Court, Sein, McCown, Hackett & Parkin, 1988, Rovet, Ehrlich & Hoppe, 1988; Varni, Babani, Wallander, Roe, Frasier, 1989; Roth & Borkenstein, 1989; Auslander, Anderson, Bubb, Jung & Santiago, 1990; Hagen, Barclay, Anderson, Feeman, Segal, Bacon & Goldstein, 1990; Rovet, Ehrlich & Czuchta, 1990; Auslander, Haire-Joshu, Rogge, & Santiago, 1990; Kovacs, Iyengar, Goldston, Stewart, Obrosky & Marsh, 1990; Jacobson, Hauser, Lavori, Wolfsdorf, Herskowitz, Milley, Bliss & Gelfland, 1990; Hauser et al., 1990; Reich, Kaspar, Puczynski, Cleland, Dell'Angela, & Emanuele, 1990; Puczynski, Puczynski, Reich, Kaspar & Emanuele, 1990; Band & Weisz, 1990; Grey, Cameron & Thurber, 1991;

Kovacs, Goldston, Obrosky & Iyengar, 1992; Weist, Finney, Barnard, Davis & Ollendick, 1993; Safyer, Hauser, Jacobson, Bliss, Herskowitz, Wolfsdorf & Wertlieb, 1993; Auslander, Bubb, Rogge & Santiago, 1993; Reid, Dubow, Carey & Dura, 1994; Kovacs, Ryan & Obrosky, 1994; Grey, Cameron, Lipman, & Thurber, 1995; Burlison Davis, Coon, Whitehead, Ryan, Burkley, & McMahon, 1995; Reid, Dubow & Carey, 1995; Simell, Moren, Keltikangas-Jarvinen, Hakalax, & Simell, 1995; Northam et al., 1996; Hanson et al., 1996; Sterling & Friedman, 1996; Rovet, 1997) (see Table 2.2 in Appendix 1). More than half of the selected studies included the physical functioning of children with diabetes mellitus or the emotional consequences. Twelve of the studies examined the cognitive functioning of children with diabetes mellitus and only two articles were directed towards social functioning.

Most of the studies including *physical functioning* looked at disease-specific complaints, diabetic care and metabolic control. Children with an early onset of diabetes (Rovet, Ehrlich & Hoppe, 1988), especially girls (Rovet, Ehrlich & Hoppe, 1987), had more hypoglycemic seizures than other groups. It was found that 30% of the children between 8 and 14 years old had serious noncompliance with the medical regimen (Kovacs, 1992), although in general, adolescents are reported to be more noncompliant than children (Johnson, Silverstein, Rosenbloom, Carter & Cunningham, 1986; Jacobson et al., 1990; Grey, Cameron & Thurber, 1991; Kovacs, Goldston, Obrosky & Iyengar, 1992; Weist, Finney, Barnard, Davis & Ollendick, 1993). In addition, it was found that with increasing duration self-management tasks were harder to do (Kovacs, Iyengar, Goldston, Stewart, Obrosky & Marsh, 1990). Weist, Finney, Barnard, Davis and Ollendick (1993) found that older children had more knowledge than younger ones with nonoptimally controlled youths having a higher knowledge than optimally controlled youths. More optimally controlled children had more controlling families. Reid, Dubow, Carey and Dura (1994) found no significant relation between knowledge and adherence when controlling for age and duration. Avoidance coping, depression and investing in close friends were related to worsened metabolic control (Grey, Cameron & Thurber, 1991). Poor metabolic control was further associated with higher levels of family stress and lower levels of family resources, psychosocial functioning and SES factors (Auslander, Haire-Joshu, Rogge, & Santiago, 1990; Auslander, Anderson, Bubb, Jung & Santiago, 1993; Jacobson et al., 1990; Grey, Cameron & Thurber, 1991; Weist, Finney, Barnard, Davis & Ollendick, 1993; Reid, Dubow, Carey & Dura, 1994; Burlison Davis, Coon, Whitehead, Ryan, Burkley, & McMahon, 1995, Rovet & Alvarez, 1997). No significant differences between optimally and nonoptimally controlled youths were found on measures of anxiety, child behavior and coping style by Weist, Finney, Barnard, Davis and Ollendick (1993). According to Gross, Delcher, Snitzer, Bianchi, and Epstein (1985) there were no significant correlations between HbA_{1c}-measures and measures of locus of control and self concept.

Initial assessment of coping and adjustment (Jacobson et al., 1990) were predictive for adherence over four years whereas family support was predictive for adherence in the short-term as well as long-term (Hauser et al., 1990). Social competence, self-esteem, aspects of family functioning, initial psychiatric status did not predict noncompliance in a study by Kovacs, Goldston, Obrosky and Iyengar (1992), whereas noncompliance was associated with psychiatric disorder later. Although the daily management tasks are invasive, children, between 8 to 13 years of age, with diabetes mellitus were more comfortable with diabetes care than previously thought (Kovacs, Brent, Steinberg, Paulauskas & Reid, 1986).

In respect of *social functioning*, Grey, Cameron, Lipman, and Thurber (1995) indicated that although there were no differences in psychosocial status in children with diabetes mellitus one year after diagnosis compared to a group of healthy children, two years after diagnosis withdrawal was higher than in healthy peers. In addition, girls with early onset diabetes had a lower social competence than boys or sibling controls and boys with late onset diabetes mellitus had more difficulties in social relations (Rovet, Ehrlich & Hoppe, 1987).

In general, average *cognitive functioning* for children with diabetes was reported, although disease variables may influence the child's cognitive functioning. Children with early onset (before the age of six or seven years old) and long disease duration (more than five years) had significantly lower IQ scores, a slower response time, reading and memory impairment, measures of visuospatial ability, memory, motor speed, attention and eye-hand coordination than healthy children (Ryan, Longstreet & Morrow, 1985; Holmes & Richman, 1985; Rovet, Ehrlich & Czuchta, 1990; Rovet & Alvarez, 1997). Spatial ability and arithmetic skills were associated with illness factors (duration of illness, age of onset, hypoglycemic convulsions, age) and many early onset children experienced difficulties at school (Rovet, Ehrlich & Hoppe, 1988). Mild hypoglycemia reduced motor performance attention and memory even after physical symptoms have subsided, the effects of a hypo centered on immediate effects rather than long-term effects (Reich, Kaspar, Puczynski, Cleland, Dell'Angela, & Emanuele, 1990; Puczynski, Puczynski, Reich, Kaspar & Emanuele, 1990).

Long-term effects after 8 years of having diabetes have also been reported, as youths performed within average range on a test of verbal memory, but the level of verbal association was lower than expected and short term memory was mildly impaired (Kovacs, Ryan & Obrosky, 1994). A study by Rovet and Alvarez (1997) revealed that children with diabetes have few problems with attention compared to healthy children and differences were only revealed in the select component of attention. Having had hypoglycemia and having a lower verbal IQ correlated with greater difficulty in selecting, focusing and inhibiting attentional components, whereas higher blood glucose levels were associated with a less adequate ability to inhibit impulsive responses (Rovet & Alvarez, 1997). Early onset children are at greater risk

for intellectual impairment particularly in their skills in the visuospatial area of cognitive processing and their mathematical ability and girls appeared to be somewhat more affected than boys (Rovet, Ehrlich & Hoppe, 1987; Rovet, Ehrlich & Hoppe, 1988). School difficulties were reflected in subscale performance and the need for remedial education services, poor attention span and difficulty in completing tasks (Roth, 1989). Simell, Moren, Keltikangas-Jarvinen, Hakalax and Simell (1995) concluded that children were doing better at school after one year post-diagnosis compared to their performance at time of diagnosis.

In general, findings indicate no maladjustment on measures of *emotional functioning*. For instance, there were no differences between children with diabetes mellitus and healthy children on locus of control or self concept (Gross, Delcher, Snitzer, Bianchi, & Epstein, 1985). In newly diagnosed children, few emotional symptoms and good self-esteem were found whereas in longer term, symptoms of depression and anxiety decreased and self-esteem increased (Kovacs, Brent, Steinberg, Paulauskas, & Reid, 1986). These results were confirmed by Northam (1996) who found that whilst at onset children scored above norm for CBCL norms, one year later their scores were significantly below these norms. Newly diagnosed children saw themselves as well-adjusted in anxiety, depression and self-esteem as healthy children. Children with higher depression scores remained however fairly stable whereas children with lower depression scores became somewhat more symptomatic over time (Kovacs, Iyengar, Goldston, Stewart, Obrosky & Marsh, 1990). A slight trend towards psychopathology was found on the CBCL although relative to normative population a higher self-esteem and lower anxiety was found (Burlison Davis, Coon, Whitehead, Ryan, Burkley, & McMahon, 1995). A study by Varni, Babani, Wallander, Roe, Frasier, (1989) found however that 25% of the children with diabetes were rated as disturbed by their parents whereas 14% of the healthy children were rated as disturbed. Looking at disease-specific outcomes more than two-thirds of the families of children with diabetes had diabetes-related fears (Simell, Moren, Keltikangas-Jarvinen, Hakalax, & Simell, 1995).

Mediating variables may be of importance. Although in general, early onset children had more behavior problems, late onset boys were also found to have an increased incidence of behavior problems (Rovet, Ehrlich & Hoppe, 1987). Whilst levels of anxiety increased in girls, they decreased in boys (Kovacs, Iyengar, Goldston, Stewart, Obrosky & Marsh, 1990). Preadolescents were less depressed, less anxious, coped in more positive ways and had fewer adjustment problems than adolescents (Grey, Cameron & Thurber, 1991). Lower levels of avoidance coping were related to more positive outcomes (Reid, Dubow & Carey, 1995). For children, both family support and peer social support were significant predictors for internalizing and externalizing behavior problems (Varni, Babani, Wallander, Roe, Frasier, 1989).

In conclusion, no studies were found indicating that children with diabetes mellitus suffer from more general physical complaints or physical limitations than healthy children. Short-term complications, hypoglycemias and hyperglycemias, are important illness-related complaints especially in older children and children with early onset diabetes. Most studies assessing physical functioning were dedicated to metabolic control and self-management. Although mediating variables are important, adherence was reported to be difficult, especially with increasing age. In relation to social functioning, just two studies were found, indicating more withdrawal and more difficulties in social relationships. Although children with early onset diabetes are reported to be at risk for developing cognitive problems, no difficulties were noted for the group as a whole, nor were there any indications found for problems on emotional functioning.

2.9 THE FUNCTIONING OF CHILDREN WITH EPILEPSY COMPARED TO HEALTHY CHILDREN

Following the selection criteria described above, 18 articles on children with epilepsy were selected (Dorenbaum, Cappelli, Keene, & McGrath, 1985; Corbett, Trimble, & Nichiol, 1985; Rodin, Schmaltz & Twitty, 1986; Noda, Schimizu, Kurachi, Murakami, Yamatani & Konishi, 1989; Blennow, Heijbel, Sandstedt, & Tonnyby, 1990; Hoare & Kerley, 1991, Suurmeyer, 1991; Austin, Risinger, Beckett, 1992; Huberty, Austin, Risinger, & McNelis, 1992a; Huberty, Austin, Risinger, & McNelis, 1992b; Hoare, 1993; Jambaque, Dellatolas, Dulac, Ponsot, & Signoret, 1993; Aldenkamp et al., 1993, Aldenkamp, 1994; Mitchell, Scheier & Baker, 1994; Schmidt, Darius, Roth, Meyer, Kroker, Wien & Brett, 1995; Carlton-Ford, Miller, Brown, Nealeigh & Jennings, 1995, Williams & Haut, 1995) (see Table 2.3 in Appendix 1). Most studies examined cognitive functioning. Eight of the selected studies included physical functioning. Seven studies included emotional functioning. Only two studies included social functioning.

Seizure frequency was often used as a predictor in the studies, which included *physical functioning*. It is reported that, in the long term, between half and two-thirds of children become seizure free (Suurmeyer, 1991; Rodin, Schmaltz & Twitty, 1986). Seizure history was the best predictor of seizure control and general health status (Mitchell, Scheier & Baker, 1994). In addition, children who continued having seizures had a statistically significant decrease in performance IQ whereas it was stable for patients in remission (Rodin, Schmaltz & Twitty, 1986). Parents were concerned about the risks associated with a seizure and the adverse effects of drug treatment on behavior, mood and alertness (Hoare, 1993). When treatment was stopped and children were cured, youths reported a cessation of complaints such as tiredness (Aldenkamp, 1994).

Children with epilepsy lost more days to illness in the year prior to the survey than the general population. Adults perceive children with a history of epilepsy as more

likely to have a life-threatening illness, and in poorer general health and more clumsy than healthy children or children with no history of epilepsy (Carlton-Ford, Miller, Brown, Nealeigh & Jennings, 1995).

Only two of the selected studies included *social functioning* (Dorenbaum, Cappelli, Keene, & McGrath, 1985; Hoare & Kerley, 1991), despite the indication that children with epilepsy were at greatest risk of maladjustment within social functioning according to Dorenbaum, Cappelli, Keene and McGrath (1985). Children were regarded by their parents as more solitary rather than destructive or acting out (Hoare, 1991).

In most of the studies assessing *cognitive functioning* academic difficulties were noticed. Children with epilepsy had increased problems with both attention and memory (Williams & Haut, 1995) and short-term memory was decreased for all children treated for epilepsy (Blennow, Heijbel, Sandstedt, & Tonnby, 1990). Epilepsy contributed to memory deficits in children not in daily life but in academic achievements (Jambaque, Dellatolas, Dulac, Ponsot, & Signoret, 1993). Nearly 40% of the children with epilepsy stayed back a class at least once before sixth grade (Huberty, Austin, Risinger, & McNelis, 1992a). As children reached adolescence most of their problems were related to academic performance, special classes and school failures (Dorenbaum, Cappelli, Keene, & McGrath, 1985). In those children whose seizures continued after 5 years, there was a significant decrease in performance IQ whereas it was stable or increased for children in remission. Decreased IQ, however, indicated a slower mental growth rather than a loss (Rodin, Schmaltz & Twitty, 1986).

Noda, Schimizu, Kurachi, Murakami, Yamatani and Konishi (1989) reported that age at onset appeared to be a factor that places the child at risk of cognitive dysfunction. Huberty, Austin, Risinger and McNelis (1992, a,b) noted however that seizure frequency, but not age at onset, seemed to be related to academic and affective problems although academic functioning was within normal range.

There was a significant association between anticonvulsant therapy and cognitive deterioration reported by Corbett, Trimble and Nichiol (1985), whereas Jambaque, Dellatolas, Dulac, Ponsot, and Signoret (1993) noticed that anticonvulsant drugs, seizure frequency and age at onset did not individually influence memory whereas type of epilepsy did. Drug withdrawal had a significant effect on psychomotor speed in a study by Aldenkamp et al. (1993). Differences, both before as well as after drug withdrawal, were also noted between children with epilepsy and healthy children, in respect of information processing and recognition of words (Aldenkamp et al., 1993). According to reports of parents, there was a correlation between Valproat therapy and the development of more pronounced attention and concentration disturbances (Schmidt, Darius, Roth, Meyer, Kroker, Wien & Brett, 1995). Polytherapy as well as epilepsy of more than 3-year duration impaired visual short-term memory but not the

auditory short-term memory or IQ. Reassessment after at least five years indicated that those who continued to have seizures, had a significant loss in performance IQ whereas it was stable or increased for patients in remission (Rodin, Schmaltz & Twitty, 1986). When medication was stopped and the medical condition improved, the educational performance did not show the same improvement, suggesting that factors other than the epilepsy or medication decreased the cognitive performance (Suurmeyer, 1991; Aldenkamp et al., 1993).

Although as a group, children with epilepsy did not differ from the norm group, evidence for behavioral problems and problems in *emotional functioning* in individual children existed (Dorenbaum, Cappelli, Keene, & McGrath, 1985). Children with epilepsy were predominantly worried, fearful, miserable and solitary rather than acting out or destructive compared to a healthy norm group, no differences were however found on self-esteem (Hoare & Kerley, 1991). A study in an open population by Carlton-Ford, Miller, Brown, Nealeigh and Jennings (1995) revealed that children with any history of epilepsy displayed more home behavior problems, depressed mood and impulsiveness than children without any history of epilepsy. No differences were however found on school behavior problems. Even when all treatment was stopped, parents tended to report epilepsy-related problems, remained worried and reported new complaints such as aggressiveness, whereas youths showed abrupt adaptation, a superpositive quality of life and a cessation of complaints (Aldenkamp, 1994).

Mediating variables are of importance. Seizure frequency was reported to be related to affective problems by Huberty, Austin, Risinger, and McNelis (1992b). There was an association between specific behavioral problems and focal cerebral dysfunction, however no association was found between medication and overall behavioral deviance (Corbett, Trimble, & Nichiol, 1985). Seizure disorder associated with a diffuse or multifunctional brain insult can produce problems in achievement and school adaptation (Huberty, Austin, Risinger, & McNelis, 1992b). Female gender, family stress, family mastery, extended family social support, and seizure frequency all predicted behavior problems (Austin, Risinger, Beckett, 1992).

In conclusion, no studies were found indicating physical limitations and social problems among children with epilepsy. Whereas social problems were noticed, only two studies included social functioning indicating more problems. Cognitive functioning was the area most often studied, revealing cognitive difficulties. Emotional problems as well as behavioral problems have been noted more often for children with epilepsy compared to healthy children.

2.10 THE FUNCTIONING OF CHILDREN WITH JUVENILE CHRONIC ARTHRITIS COMPARED TO HEALTHY CHILDREN

Following the selection criteria described above, 18 articles on children with juvenile chronic arthritis were selected (Billings, Moos, Miler & Gottlieb, 1987; Thompson, Varni & Hanson, 1987; Daniels, Moos, Billings & Miller, 1987; Varni, Wilcox & Hanson, 1988; Ungerer, Horgen, Chaitow, & Champion, 1988; Konkol, Lineberry, Gottlieb & Shellbie, 1989; Vandvik, 1990; Ennett, DeVellis, Earp & Kredich, 1991; Harris, Newcomb & DeWanter, 1991; Timko, Stovel, Moos & Miller, 1992; Daltroy, Larson, Eaton & Partridge, 1992; Berry, Hayford, Ross, Pachman, & Lavigne, 1993; Doherty, Yanni, Conroy & Breshnihan, 1993; Timko, Baumgartner, Moos & Miller, 1993; Ross, Lavigne, Hayford & Berry, 1993; Baildam, Holt, Conway & Morton, 1995; Hagglund, Schopp, Alberts, Cassidy & Frank, 1995; Malleson, Bennett, MacKinnon, Jespresen, Coutts, Turner & McKenzie, 1996) (see Table 2.4 in Appendix 1).

Most studies looked at *physical functioning*. In general, physical complaints and limitations are reported in children with juvenile chronic arthritis. Somatic complaints were higher than in healthy children (Thompson, Varni & Hanson, 1987; Konkol, Lineberry, Gottlieb & Shellbie, 1989). Mothers indicated diminished athletic competence (Ennett, DeVellis, Earp & Kredich, 1991). Disease severity was found to be related to athletic competence and perceptions of physical attractiveness (Ennett, DeVellis, Earp & Kredich, 1991; Varni, Wilcox & Hanson, 1988) or physical problems (Billings, Moos, Miler & Gottlieb, 1987). Remarkable results were revealed by Malleson, Bennett et al., 1996, as there were no significant differences found between children with juvenile chronic arthritis and controls for either aerobic or anaerobic fitness, only a small minority rated athletic competence lower than healthy controls. However, severity or persistence of arthritis over time might play a role in this study.

Mediating variables are important. For instance, older children seemed to experience more pain (Timko, Baumgartner, Moos & Miller, 1993; Hagglund, Schopp, Alberts, Cassidy & Frank, 1995), but pain was reported less frequently the longer children had juvenile chronic arthritis (Hagglund, Schopp, Alberts, Cassidy & Frank, 1995). Father's initial depression was predictive for children having more functional disability (Timko, Baumgartner, Moos & Miller, 1993). Although pain and somatic complaints were increased among children with juvenile chronic arthritis, family environment and child psychological factors were found to interact with disease-specific parameters to modulate pain experience (Thompson, Varni & Hanson, 1987). Child psychological adjustment, family psychosocial environment and pain in combination with disease activity statistically predicted functional status (Varni, 1988). Child's distress, maternal distress and family harmony or conflict had high correlations with children's reported pain (Ross, Lavigne, Hayford & Berry, 1993). No correlations between psychological measurements and functional measures were however found by Baildam, Holt, Conway

& Morton (1995). No studies were found reporting on daily management such as having medication or using special aides.

Eight of the selected studies included *social functioning*. Social competence difficulties were found among children with juvenile chronic arthritis (Thompson, Varni & Hanson, 1987). Whereas social problems occurred slightly more frequently, older age and shorter duration as well as severity were correlated with poorer social competence (Daltroy, Larson, Eaton & Partridge, 1992; Ennett, DeVellis, Earp & Kredich, 1991). More social activities of the mother correlated with more social competence among patients. Disability and father's personal strain predicted less social competence of the children (Timko, Baumgartner, Moos & Miller, 1993). In addition, a lower self concept was related to feeling more lonely and being more teased and having less to do in leisure time (Ungerer, Horgen, Chaitow, & Champion, 1988).

Only four of the selected studies included *cognitive functioning*. It was found that children with juvenile chronic arthritis had a higher absenteeism score than other children (Billings, Moos, Miler & Gottlieb, 1987) and were concerned about school (Konkol, Lineberry, Gottlieb & Shellbie, 1989). The severe group and older severe patients were more likely to miss school (Berry, Hayford, Ross, Pachman, & Lavigne, 1993), and functional disability correlated with poorer school grades (Timko, Stovel, Moos & Miller, 1992). No reports of cognitive dysfunction or school failures were however found by Berry, Hayford, Ross, Pachman and Lavigne (1993). In respect of the understanding of illness, the conceptual level was a better predictor for their understanding of illness than age.

Fourteen studies included *emotional functioning*. Psychosocial problems of at least mild severity was found in 64% of the children with juvenile chronic arthritis, but the total behavior scores were close to American and Dutch norms in healthy children. Levels of competence, adjustment and stress were comparable to those of healthy controls (Vandvik, 1990). Although self concept (Malleison, Bennett et al., 1996; Baildam, Holt, Conway & Morton, 1995) and depression levels were comparable to a healthy population, psychopathology was found in 21% of the children with juvenile chronic arthritis in comparison with 13% in healthy controls (Baildam, Holt, Conway & Morton, 1995). Disease-specific measures revealed that one third of the children had a negative attitude towards juvenile chronic arthritis (Doherty, Yanni, Conroy & Breshnihan, 1993). In addition, children with arthritis and their siblings reported to be scared of death (Konkol, Lineberry, Gottlieb & Shellbie, 1989).

Subjects in the severe group had more parent-reported psychological problems than mild patients or healthy controls (Billings, Moos, Miler & Gottlieb, 1987) and a lower self concept (Ungerer, Horgen, Chaitow, & Champion, 1988). No correlation between psychological scores and functional measurements were however found by Baildam,

Holt, Conway and Morton (1995). Higher parental depression, medical symptoms, family symptoms and burden of illness predicted more problems (Daniels, Moos, Billings & Miller, 1987). Parental distress was correlated with children's behavior problems (Daltroy, Larson, Eaton & Partridge, 1992). Concurrent rather than prior risk, resistance and disease related factors predicted psychosocial outcome (Timko, Stovel, Moos & Miller, 1992). Girls had similar scores to the healthy population, boys showed a slight trend towards behavior problems and the oldest children had the most behavior problems (Daltroy, Larson, Eaton & Partridge, 1992). Poor baseline functioning was a strong predictor for functioning 4 years later (Timko, Baumgartner, Moos & Miller, 1993).

In conclusion, physical complaints and limitations were more often reported in children with juvenile chronic arthritis compared to healthy children. Children with juvenile chronic arthritis are reported to have a higher absenteeism score than healthy children, while their academic achievements are reported to be comparable to healthy children. Severity of illness may however play a role in this. There are no conclusive results with regard to the emotional functioning of children with juvenile chronic arthritis.

2.11 STUDIES AMONG DIFFERENT DIAGNOSTIC GROUPS

Although most of the studies found, examined the functioning of children with a chronic illness in comparison to healthy children, 22 studies were found in which different diagnostic groups were used. As described in Chapter 1, studies revealed that most children with a chronic illness do well, although psychosocial problems occur less often among healthy children (Perrin, Ramsey & Sandler, 1987). Children with a chronic illness are reported to have more behavioral and social competence problems than the healthy norm group, although the adjustment is better than that of a normative sample referred to mental health clinics. Most prevalent problems are social competence problems (20%) and internalizing problems (12%) (Wallander, Varni, Babani & Banis, 1988). On the other hand, children with a chronic illness are also reported to have significantly higher levels of empathy and emotional responsiveness than healthy children, behaviors that may be facilitated by illness experiences (Nelms, 1989). Apart from studying differences between healthy children and children with a chronic illness as a group, studies examined differences between disease specific groups.

Children with asthma were reported to have more difficulties in taking care of themselves than children with diabetes mellitus or healthy children (Sillanpaa, 1987). Despite the fact that neurological impairments are viewed as being more disabling and handicapping than non-neurological ones, more mobility and occupation handicaps in children with asthma were reported (Sillanpaa, 1987). Children with asthma had a

greater functional impairment than children of other groups (Padur et al., 1995) and a significantly lower quality of life on all physical domain variables compared to children with epilepsy (Austin, Smith, Risinger, & McNelis, 1994). Although children with epilepsy had more behavior problems, most mean scores on behavior problems (especially home behavior problems) for children with asthma were also above mean scores of the general population (Austin, 1989). Mothers of children with asthma reported a greater number of internalizing behavior problems than mothers of children with diabetes mellitus did, although the mean score was below clinical significance (Hamlett, Pellegrini & Katz, 1992). Children with asthma had a lower self concept score than healthy children or children with diabetes mellitus (Nelms, 1989). More depression and a lower self concept was noted for children with asthma compared to children with cancer, diabetes mellitus or healthy children (Padur, 1995).

Children with diabetes mellitus more often take part in leisure activities than other impaired children (Sillanpaa, 1987). Children with diabetes mellitus had a lower sense of school competence than children with a recent acute medical problem, but other between-group differences were not found by Jacobson et al. (1986). The results on the Diabetes Adjustment Scale showed that children were not consciously overwhelmed by onset of diabetes. Differences between groups on levels of internalizing and externalizing child behavior symptoms were nonsignificant when social class was controlled (Wertlieb, Hauser & Jacobson, 1986).

Social competence of children with diabetes mellitus and asthma does not differ from that of healthy peers on measures of social performance and skills (Nassau & Drotar, 1995). In contrast, children with diabetes mellitus scored high on self-reported aggression, compared to healthy children and children with asthma (Nelms, 1989). Children with diabetes mellitus displayed lower scholastic self-competence than children with asthma (Holden, Chmielewski, Nelson & Kager, 1997).

Children with epilepsy had significantly more behavior problems at school and at home, more inattentiveness, more anxiety, more social withdrawal, and were less popular than children with asthma (Austin, 1989). In addition, both children with epilepsy and children with asthma had a heightened level of difficulties in peer relations compared to children with diabetes, cardiac disease, leukemia according to Eiser (1992). In addition, mothers reported children with epilepsy as having more difficulties with frustration and hostility feelings than children with diabetes, asthma, cardiac disease and leukemia. Children with petit mal seizures were less well-adjusted according to teachers than children with other illnesses (Perrin, Ayoub & Willett, 1993) and more behaviorally disturbed, having a lower self esteem than children with diabetes (Hoare & Mann, 1994). Children with epilepsy had more emotional and behavioral problems as well as more problems in the social and

school domains and had a more compromised quality of life than children with asthma (Austin, 1994).

Children with juvenile chronic arthritis displayed fewer externalizing behavior problems than children with another chronic illness (Wallander, 1989). Children with juvenile chronic arthritis respond with cognitive coping strategies as often as healthy children or children with another chronic illness (Olson, Johansen, Power & Pope, 1993).

2.12 CONCLUSION

Children with asthma, diabetes mellitus, epilepsy or juvenile chronic arthritis, have to cope with physical complaints, limitations and self-management tasks. The symptoms and treatment of these illnesses have similarities but also unique characteristics that have to be taken into account when studying the consequences of the illnesses. All children have to cope with symptoms or complaints, daily treatment tasks or medical visits. As such, having a chronic illness, irrespective of diagnosis, has an impact on health-related quality of life. In addition, children have to cope with disease specific characteristics. Children with asthma or children with epilepsy may experience attacks or seizures that may occur unexpectedly, whereas children with diabetes may experience hypoglycemias and hyperglycemias. Children with juvenile chronic arthritis may experience symptoms that vary in severity over time, such as pain, or swollen joints. Where with diabetes mellitus, a diet and insulin injections are of importance, with juvenile chronic arthritis, rest, physical therapy and special aides may be necessary. Children with asthma and epilepsy have to take their medication. All children need thus to take their illness into account in daily life.

A review of clinical studies among these groups revealed that, although it has been reported that 20% suffer from problems in functioning, the consequences of childhood chronic illness differ widely, depending on mediating variables. Age, gender, illness characteristics or severity, family variables and internal resources have often been included in studies assessing the impact of illness. General conclusions are therefore difficult to formulate. In addition, it must be noted that in this review no distinction was made between groups within one disease category. Epilepsy seizures are very distinct from each other and may therefore have a distinctive impact on the child's life. The same is true for children with distinct forms of juvenile chronic arthritis or for children with distinct severity of illness within one diagnostic group. In addition, it must be noted that this review was far from exhaustive. Nevertheless, general conclusions with regard to main areas of problems for each illness category may be drawn.

It was concluded that children with asthma are at increased risk of physical consequences. Physical symptoms or complaints are greater than in healthy children and asthma may lead to a reduced involvement in physical activities, although a more important factor seems to be the limitations of performance in these activities. Although results on social functioning are not conclusive, it might be concluded that children with asthma are at increased risk for problems in social functioning. Children may also experience limitations especially the degree to which they can participate in social activities rather than in their social relationships. With regard to the cognitive functioning of children with asthma, it is concluded that although children with asthma have a higher school absence than healthy children, they do not in general experience severe decrements in school performance due to their asthma. Mainly emotional or internalizing problems, rather than behavioral problems, appear to be increased among children with asthma.

No studies were found indicating that children with diabetes mellitus have more physical limitations or general physical complaints or cognitive difficulties than healthy children, except for the fact that children with early onset of diabetes, and especially early onset girls, are at risk in relation to physical functioning, cognitive functioning and emotional functioning. Physical consequences were studied mostly in relation to adherence and metabolic control. There was no examination of general physical complaints or physical limitations. Adherence and metabolic control seemed in part to be dependent on family factors and age. In the primary school age group, children are more comfortable with daily management than was previously thought. The cognitive functioning seems to be within average range, although children with early onset diabetes mellitus and children with hypoglycemic attacks are seen as at increased risk of developing cognitive difficulties at special tasks. Social problems were however more prevalent in children with diabetes mellitus than in healthy children, but only two studies were found on this issue. Studies on emotional or behavioral problems revealed, in general, no increased problems, although in one study illness-specific fears were noticed in a majority of children and families. In addition, the increased problems seem to be persistent over time, so that it is worthwhile identifying the children who are at risk.

With respect to the physical functioning of children with epilepsy, most studies examined disease-specific complaints and prognosis. The experience of daily treatment tasks was not assessed in the selected studies. Although it was suggested that children with epilepsy could be at risk in developing social problems, only a few of the studies included social functioning variables. Studies on the cognitive functioning of children with epilepsy were however prevalent. In general, results indicate that children with epilepsy are at risk of developing cognitive problems. While it is suggested that these problems are due to the illness and treatment, also other variables such as expectations from their environment are thought to play a role. In addition, it can be

concluded that children with epilepsy are at risk in developing behavioral as well as emotional disturbances.

Given the illness characteristics in juvenile chronic arthritis, such as mobility difficulties and pain, children are at increased risk of physical problems. While children with arthritis may be confronted with invasive daily treatment tasks, as special aides, rest and exercises, the experience of daily treatment tasks were not assessed in the selected studies. Although school absence may be higher for children with juvenile chronic arthritis, *no significant difficulties in school-related functioning were found*. Children with juvenile chronic arthritis, especially children with severe symptoms, are also reported to be restricted in social functioning. Cognitive and social functioning are however the least studied domains. In general, it has been suggested that children with juvenile chronic arthritis have not more or slightly more emotional or behavioral problems than other groups.

Reviewing the dimensions studied within each group, it can be concluded that expected difficulties in specific illnesses were more extensively studied than other dimensions. For all diagnostic groups, emotional consequences were studied most frequently. Most emotional problems were studied in general terms. There were only two studies which revealed disease-specific concerns of children with diabetes mellitus and juvenile chronic arthritis (Simell, Moren, Keltikangas-Jarvinen, Hakalax & Simell, 1995; Konkol, Lineberry, Gottlieb & Shellby, 1989) and one study assessing the attitudes towards illness (Austin & Huberty, 1993). Except for children with epilepsy, the physical consequences were the second most frequently studied area of difficulties. For children with epilepsy, the cognitive consequences were relatively often studied, whereas for children with juvenile chronic arthritis this was the least studied domain.

Within-group comparisons revealed that more disease-related physical complaints, as having attacks in asthma, seizures in epilepsy, hypoglycemias or hyperglycemias in diabetes mellitus or juvenile chronic arthritis related symptoms could lead to more physical, social or psychological consequences, although mediating variables, such as coping or social support, are of importance.

In most studies problems of children are assessed in comparison with a healthy group or a normative sample. Most studies used general instruments whereas the use of illness-specific scales is rare and only used in separate studies. The appropriateness of the general instrument norms or generic instruments can however be questioned since children with a chronic illness were not included in the reference group and items could reflect merely a disease symptom instead of a symptom of adjustment problems (Drotar, 1997). General instruments may include items that represent depression or anxiety in healthy children whereas in a child with an illness the same symptoms may represent normal physical consequences of the child's

medical condition. Unfortunately higher scores on such instruments may be misinterpreted as indicative of poorer adjustment (Johnson, 1991). In addition, general instruments may demonstrate a low sensitivity and high specificity when used among children with a chronic illness. An explanation for this may be that the instruments have difficulty in detecting internalizing disorders, which predominate in this group (Harris, Canning & Kelleher, 1996). It is therefore recommended that the reliability and validity of general instruments on a population of children with a chronic illness should be assessed and the applicability of items included should be checked. In addition, the generic instrument also has to include children with a chronic illness as a normgroup. This is especially true for the assessment of psychological problems for which many instruments are available and currently used among children with a chronic illness. Whilst the specific impact of diagnosis is hard to detect by general studies and the use of general measures, disease-specific measures also have to be included in research. In particular, disease-specific physical consequences as well as psychological fears and concerns related to having an illness are areas in which generic instruments do not suffice and which are seldom studied.

Very few studies included the daily management of illnesses, but a more specified literature search looking for studies on adherence or self-management should reveal far more studies on this subject. In addition, very few studies included the child's feelings related to daily management. Although these studies add up to our knowledge of functioning of children with specific diseases in dimensions where difficulties are to be expected, a holistic view on the functioning of the child is missing. Unexpected problems may thus easily be overlooked. The current focus in research as well as in clinical practice is to integrate the biological, social and psychological consequences of having a chronic illness. Pediatricians nowadays work in a team and co-operate with psychologists, general practitioners, (specialized) nurses and other health care workers. Broader health outcomes together with psychosocial measures suitable for use among children with a chronic illness are therefore needed. A modified approach, using generic as well as disease-specific measures is necessary to enhance knowledge on the consequences of illness on distinct areas of functioning.

"Health-related quality of life" is an umbrella term in which broad areas of functioning are included. In addition, the assessment of health-related quality of life may be a key step in, assisting the children and families to manage the illness and to understand the functioning and feelings of the child.

CHAPTER 3: HEALTH-RELATED QUALITY OF LIFE AMONG CHILDREN WITH A CHRONIC ILLNESS, THE DEFINITION AND ASSESSMENT

3.1 INTRODUCTION

Health-related quality of life is often used as a construct to define the broader spectrum of outcomes of illness and treatment and is becoming increasingly important in the expanding area of medical care.

The term health-related quality of life was first used in medicine in connection with patients dying or neonates who were severely handicapped. Evaluation of health-related quality of life however is now routine in all kinds of medical decisions, at least among adults and is no longer restricted to life or death situations. Health-related quality of life is especially important in chronic illnesses, where cures are still elusive and the interaction between physician and patient is a necessity (Ehrlich, 1988).

Effects of illness, side effects of treatment and choices between treatments create a need for health-related quality of life assessment. In addition, it is known that people with the same symptoms and methods of treatment can experience different levels of physical limitations and feelings of distress. The question is no longer whether these factors should be measured but what are the most reliable and practical means of obtaining these data (Slevin, Plant, Lynch, Drinkwater & Gregory, 1988).

Despite the increasing attention given to health-related quality of life among adults, the recognition that health-related quality of life assessment is of major importance in chronic illness, as well as the fact that nearly 20% of the children are chronically ill, research on health-related quality of life among children is, as yet, very limited. This is partly due to the fact that there is a shortage of suitable quality of life measures for children with a chronic illness (Mulhern et al., 1989; Rosenbaum, Cadman & Kirpalani, 1990; La Greca, 1994; Van Elderen et al., 1994). There is a pressing need for measures that adequately capture the components of health-related quality of life in children. Measuring health-related quality of life in children is no longer an optional extra - it is essential (Kind, 1994). In response, the HOW ARE YOU? (HAY), a health-related quality of life questionnaire for children of 7 to 13 years with a chronic illness was developed.

In order to be able to develop a health-related quality of life instrument and to assess the merits of the instrument, several criteria have to be established. In this chapter, these criteria will be discussed. The use of these criteria and reasons for the choices that were made will establish a solid theoretical background of the HAY.

First of all, the definition of health-related quality of life is described since quality of life is often used as an umbrella term used in the context of very diverse topics. Secondly, the purpose of health-related quality of life assessment is formulated. Depending on the aim of the study or the diagnostic research, instruments can be chosen that apply to conform to that aim.

Through consideration of the purpose of assessment and definition of health-related quality of life, the content of measurement can be established. Several questions are answered in order to compose the instrument: a) Which components of life have to be included b) Do we need a multidimensional profile or a single score? and c) Do we need a generic or disease-specific measure?

After establishing the definition, the purpose and content of the questionnaire, the question arises as to who is the source of information. When the answer includes the children themselves, the following question will be whether primary school children are able to understand and use rating scales to quantify health-related quality of life.

After formulating the answers to these questions and thereby our criteria for developing the HAY, the problem arises of which instruments are available for children with a chronic illness, especially for primary school children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis, these being the target groups in this doctoral dissertation.

3.2 SCOPE OF HEALTH-RELATED QUALITY OF LIFE

Although health-related quality of life is a relatively new concept, its modern conceptual basis originates in the WHO definition of health formulated in 1947, which defined health as “a state of complete physical, mental and social well-being and not merely the absence of disease and infirmity”. A few years later Karnofsky and Burchenal signaled the introduction of psychosocial or health-related quality of life considerations (Aaronson, 1990). In the following years health-related quality of life research was increasingly applied in clinical trials among adult patient groups. The term is however very broadly used without explicitly stating the theoretical basis. One reason for this may be that health-related quality of life measurement is a response from the clinical practice and often conducted in applied studies (Haes, Knippenbergh & Trijsburg, 1991). The consequence is that the meaning of health-related quality of life is different in distinct areas of research, even within the restricted area of health research. In order to have a proper discussion on the use of health-related quality of life measures within the area of health, and more specifically, among children with a chronic illness, it is essential to clarify the meaning of health-related quality of life and to differentiate between concepts which are often used interchangeably when considering health-related quality of life: health, health status and functional status.

Good health is our most desired and sought after state of being. Despite the central place occupied by health in our lives, health as such is not a well-defined concept. Traditional approaches in health studies include mortality rates and morbidity rates. Morbidity rates however do not reflect the impact of illness upon individuals and also ignore mortality (Kaplan, 1985). Authors use health measures to estimate health-related quality of life and use these terms interchangeably. In order to distinguish between the terms, health is here defined as the absence of physical illness. Impairment in the context of health experience is defined as any loss or abnormality of psychological, physiological, or anatomical structure or function and concerns the biological function of the body. Disability is concerned with any restriction or lack (resulting from impairment) of ability to perform an activity in the manner or within the range considered to be normal. A handicap represents the social consequences of impairments or disabilities (Bowling, 1991).

In the case of chronic illness, functional status can be defined as “the degree to which an individual is able to perform socially allocated roles free of physically related (or mentally in the case of mental illness) limitations” (Bowling, 1991, p.6). A general health measure does not need to take into account the ability to perform social roles (Bowling, 1991) whereas measures on functional status are focused on these issues. In addition, attention in health research has largely been focused on limitations, disability and functional status rather than on a personal evaluation of these issues.

Currently there is a growing awareness that health-related quality of life has to include this personal estimation of value. In addition, health-related quality of life is increasingly recognized as a multidimensional construct. Eiser, Havermans and Eiser (1994) asked a small group of healthy young adolescents (mean age 13 years) what they think “quality of life” means. The majority of children were able to define the construct. One-third gave a general definition, while a quarter gave an extra judgmental comment that included the appreciation of what’s good about life. An essential component of health-related quality of life was according to the youngsters the quality of relationships with family and friends. A significant number included their own physical health, material possessions, physical appearance and task performance in their personal definitions. Independence and future opportunities were less often noted. When asked in what way illness would affect their lives, most children answered in terms of social problems or isolation, others responded in terms of emotional problems (Eiser, Havermans & Eiser, 1994). In the following paragraphs, the two characteristics of health-related quality of life mentioned, multidimensionality and subjectivity, are delineated.

3.3 MULTIDIMENSIONALITY

Although most authors seem to agree on the multidimensional character of health-related quality of life, some prefer a single score. For example, Tofler (1995, p.444) stated "However, the complexity of current assessments of health-related quality of life lessens their usefulness in the clinical setting. I have found a simple grading, from one to five, by the patient to be quite adequate". De Haes, Knippenbergh and Trijsburg (1991) stated that it seemed logical that one asks for a judgement about life as a whole compiled of the aggregation of distinct judgements in health-related quality of life. Cunny and Perri (1991) concluded from an investigation with MOS Short-form General Health Survey that the significant correlation of the single item measure of health-related quality of life with the total score suggests that such a simple measure is an acceptable method of assessing health-related quality of life. Cunny and Perry added however that the single item cannot provide detailed information. Although a single score can be beneficial in policy decisions, in clinical areas it does not suffice (Leger, 1988). In addition, the summing of disparate dimensions is not recommended because contradictory trends for different aspects of health-related quality of life are missed (Fitzpatrick, Fletcher, Gore, Jones, Spiegelhalter & Cox, 1992a). In particular it is advocated that children's health-related quality of life should be represented in a profile as children have a greater ability than adults to overachieve in some areas and to make gains in other functions or abilities within a developmental framework (Starfield, 1987; Mulhern et al., 1989).

Multidimensional instruments allow one to disentangle the positive and negative effects of a given treatment and to facilitate the selection of the most appropriate treatment plans. In addition, health-related quality of life of patients may be affected in different ways at different stages of the disease process and treatment process, all of which cannot be identified in a single score. The third benefit of multidimensional instruments is that it is possible to document both anticipated and unexpected effects of illness and treatment (Aaronson, 1990). In addition, by measuring multidimensional, a more holistic view on important dimensions of functioning is obtained.

When considered as a multidimensional construct, health-related quality of life is often seen as reflecting functioning on three dimensions: physical functioning, social functioning and psychological (Aaronson, 1990; Joyce, 1988; Schipper, Clinch & Powell, 1990; Patrick & Erickson, 1988; Ware, 1984; Guyatt & Jaescke, 1990). Some authors also add an economic dimension (Joyce, 1988). Patrick & Erickson (1988) however excluded social, economic or cultural aspects of health-related quality of life, as the focus of clinicians and caretakers is primarily on health-related quality of life, but admitted that this can be difficult as diseases like rheumatoid arthritis can affect social functioning or economic areas of life. Whilst economic aspects may also influence children's daily life, it is mostly parents who are more involved with these

issues so that this domain does not have to be included in an instrument to measure the child's quality of life. Whereas health-related quality of life is most often oriented towards malfunctioning, Hyland (1992) stresses the importance of happiness in health-related quality of life assessments, as complaints often included in health-related quality of life assessment are likely to be independent of positive life quality.

In assessing children's health-related quality of life, the dimensions have to be related to children's daily life and feelings. As there are large differences in the meaning of health between children and adults and even between children of various ages, different operationalizations for distinct age groups are necessary. The concept of health in pre-scholars is for example different from health in primary school children and this is also different from the concept of health in adolescence (Starfield, 1987). The dimensions of functioning applicable to adults may therefore not be directly relevant to a child's functional status and other dimensions also need reformulation (Rosenbaum, Cadman & Kirpalani, 1990). The challenge is to find concepts that guarantee the assessment of the relevant issues in children with a chronic illness (see Chapter 2).

3.4 SUBJECTIVITY

As described in Chapter 1, health-related quality of life is dependent on other life events, coping abilities, internal and external resources but also on values and goals of patients. health-related quality of life thus encompasses the individual responses to the physical, social and psychological consequences of illness and should therefore only be described in individual terms. Currently, most authors agree more or less on the subjective nature of health-related quality of life.

According to de Haes, Knippenbergh and Trijsburg (1991), health-related quality of life is a judgement of the patient of the good and satisfying character of life. According to the authors, this attitude should be measured by a procedure that locates the subject on a bipolar affective or evaluative dimension vis-à-vis a given object. Others suggest including an individual dimension in health-related quality of life instruments, a dimension on spiritual life or environment, things that really matter to the patient. As a baseline, an individual health-related quality of life score which varies from patient to patient can be used to assess changes over time and allow for a greater sensitivity for change (Joyce, 1995; Fitzpatrick et al., 1992).

Calman (1984) was one of the first authors to define health-related quality of life as the discrepancy at a particular time between the hopes and expectations of the individual and the individual's present experience. This definition is concerned with the difference between perceived goals and actual achievements (Calman, 1984). Related to this conception, Patrick and Erickson (1988) used weightings to identify the

health-related quality of life of a person and defined health-related quality of life as the level of well-being and satisfaction associated with events or conditions in a person's life as influenced by disease accidents or treatments. Thus, it is not only the ability to perform social roles but also the degree of satisfaction derived from performing them which constitutes health-related quality of life (Patrick & Erickson, 1988). In the Ferrans and Powers Qol Index patients are asked to rate how satisfied they are with aspects of their life and then rate how important the aspects are to them. In this way, scores reflect how satisfied people are with things that matter most to them (Ferrans, 1990). In trying to formulate a theory of health-related quality of life, Nordenfelt (1995) constructed a theory according to which health-related quality of life is a property pertaining to the experiences of the individual. Positive health-related quality of life is defined by the emotion of happiness and the sensation of pleasure in which happiness is an equilibrium between the subject's wants and the world he or she finds it to be. Felce and Perry (1995) proposed a model of health-related quality of life that integrates objective and subjective indicators of a broad range of life domains and individual values. In their model, health-related quality of life is defined as an overall general well-being that comprises objective descriptors and subjective evaluations of physical, material, social and emotional well-being together with the extent of personal development and purposeful activity, all weighted by a personal set of values and aspirations. All three sets of variables, objective descriptors, subjective evaluations and personal values, have to be measured in their view.

These conceptualizations lead to the need for an assessment incorporating the (objective) situation as well as the value people attach to this situation. In this way, help can be directed to improving the situation or to redirecting the hopes and expectations and thereby decreasing the gap between functioning and evaluations. When the evaluation is positive, the gap between expectations and achievements is small. When the evaluation is negative, a gap exists between achievements and expectations and help is needed. While few measures include the patient's perceptions of improvement or satisfaction with level of performance, these perceptions are predictive for individuals seeking care, accepting treatment and considering themselves to be well (Bowling, 1991).

3.5 DEFINITION OF CHILDHOOD HEALTH-RELATED QUALITY OF LIFE

In accordance with the discussion described above, health-related quality of life is defined as a multi-dimensional concept in which the level of performance as well as the personal evaluation by the child is of great importance. The following definition will serve as the basic precept in the development of the HAY: "Health-related quality of life in children with a chronic illness refers to the physical, social and psychological functioning of children, and the gap between their achievements and

expectations related to their functioning. The functioning of children is described in terms of prevalence of activities, symptoms, problems or emotions and the quality of performance in activities. The gap between achievements and expectations is defined by the appraisal of difficulties related to the physical, social and psychological functioning". To decrease the gap, treatment can be directed toward achievements as well as expectations so that these have to be estimated separately. Since achievements as well as expectations may differ depending on, for instance, changes in illness and treatment characteristics, time, other events, internal and external resources, values and coping abilities, health-related quality of life is a dynamic construct and has to be measured over time.

3.6 PURPOSES OF HEALTH-RELATED QUALITY OF LIFE ASSESSMENT IN CHILDHOOD

Justification for health-related quality of life measurement among children can be provided at a number of levels. In clinical practice, health-related quality of life instruments can be useful for they sometimes uncover symptoms or restrictions in activities which are important and possibly remediable but which patients fail to mention or of which physicians might not be aware (Patrick & Erickson, 1988). In a pediatric population this is even more important than among adults, as children often use a different vocabulary to physicians and are possibly not used to mention symptoms or restrictions by themselves. In addition, physicians may not be used to asking children to reveal complaints and ask thus their parents. Valid and reliable measures of health-related quality of life permit the physician to assess more objectively the outcomes of health interventions of all kinds, which is especially important because physicians are inaccurate judges of patient's functioning and health-related quality of life (Sturner, 1991). Describing the nature and extent of problems confronting the patient facilitates a shared view of impact of illness and treatment (Fitzpatrick et al., 1992). Health-related quality of life instruments will thus assist in improving the communication between physician and patient (McGee, 1994). Furthermore, health-related quality of life instruments can assist in screening individual patients for the need for additional medical or psychosocial interventions (Fitzpatrick et al., 1992; Jaeschke & Guyatt, 1990). They may also be used as evaluative instruments to be able to distinguish between those effects which are positive and those which are not (Kind, 1994).

A few cautions should be aired as the children or their parents must realize that questionnaires are only a way of listing their own concerns. In addition, questionnaires do not generally have sufficient validity to be considered alone, so that the data should be considered within the context of other data. Yet, pediatricians should value questionnaires as timesaving adjuncts and appreciate how they help to focus attention on problem areas and on the resulting improved medical record (Sturner, 1991; Greer, 1987). Although health-related quality of life instruments are

not intended to replace clinical judgement, they can assist in structuring and transmitting clinical information and thus enhancing its credibility. A comment often voiced, however, is that health-related quality of life data are “soft” rather than “hard” data. Yet, despite pretensions to the contrary, most medicine is based on soft data, derived from patient and physician perception (Greer, 1987).

At the research level, health-related quality of life measurement can assist in screening pediatric populations at risk. In addition, population surveys of perceived health problems may establish norms for psychosocial morbidity among specific patient groups (Leger, 1988). Furthermore, health-related quality of life assessment may assist in assessing the impact of medical treatment on the child’s health-related quality of life and permit differentiation between medications or interventions (Leger, 1988; Spilker, 1990; Fitzpatrick et al., 1992). After publishing the results, data may be used clinically in prescribing medication or intervention or commercially in advertising the results.

At policy level, an important use of health-related quality of life data may be improving the allocation of health care resources (Spilker, 1990). Furthermore, health-related quality of life assessment can assist in cost-utility analyses (Fitzpatrick et al., 1992).

3.7 GENERIC AND DISEASE-SPECIFIC METHODS

In health-related quality of life research there has been a debate on whether to use generic or disease-specific measures. In the past, there has been a tendency to use generic measures, designed to summarize a spectrum of the concepts of health-related quality of life that apply to many impairments. Generic measures were readily available and it was presumed that general functioning would predict disease-specific difficulties, for instance general anxiety was presumed to predict anxiety concerned with the illness (Johnson, 1991). An advantage of generic measures is that they can be applied to all patient groups irrespective of their diagnosis (Patrick & Deyo, 1989; Guyatt & Jaeschke, 1990).

Generic measures are helpful in deciding on differential benefits, and in addressing difficult questions surrounding the priorities in health care. In addition, these measures can generate important information about the impact of treatment on the patient as a whole. Generic measures enable observations to be made on several aspects simultaneously (Kind, 1994). In addition, the use of generic instruments will permit the comparison of different populations and different programs, an important objective for policy analysis and decision-making (Patrick & Ericksson, 1988; Aaronson, 1990). The benefits of a generic measure are thus that only one instrument is needed among distinct groups and that it allows for comparisons between groups,

interventions or conditions. Furthermore, when the general functioning of the patient is being examined, then generic measures are appropriate (Johnson, 1991).

Two disadvantages of generic measures are, however, that they may not adequately address issues of particular relevance in evaluating the effect of a specific disease or treatment on health-related quality of life and may not be sufficiently responsive (Aaronson, 1990; Jambon & Johnson, 1997).

Disease-specific measures are measures designed to assess specific diagnostic groups or patient populations and range from those designed for a special group to those designed for use among a broad spectrum of related diagnostic categories (Patrick & Deyo, 1989; Aaronson, 1990; Guyatt & Jaeschke, 1990). The last category offers the opportunity of cross-study comparison, while still providing a relatively targeted assessment of health-related quality of life issues relevant to the study group (Aaronson, 1990). The detailed data will contribute most significantly toward better treatment planning at the level of the individual patient. A disease-specific measure has thus much in common with clinical measures. Most clinicians feel that some disease-specific questions are necessary (Leger, 1988). The addition of disease-specific supplements to generic measures to assess special concerns will probably increase the acceptance of measures by patients and specialists (Patrick & Erickson, 1988). In addition, the inclusion of disease-specific issues, relevant to target groups, is an essential requirement to increase the sensitivity to change of the measure (Fitzpatrick et al., 1992). In this doctoral dissertation, chronic illness measures are considered as measures for children with a chronic illness irrespective of their diagnosis, whereas disease-specific measures are meant for specific patient populations. The dual approach, considering generic as well as disease-specific aspects, is one step towards broadening the narrow approach taken in providing health care to children (Lewis, Pantell & Kieckhefer, 1989) and is consistent with the modified categorical approach.

3.8 SOURCE OF INFORMATION

As health-related quality of life is subjective and related to the experience and feelings of an individual person, the only realistic reference for use in the judgement of health-related quality of life is the patient him- or herself. There is a growing consensus on the issue, at least among adult patient care, that the individual him/her self is the only proper judge of his health-related quality of life (Sprangers & Aaronson, 1992; Ferrans, 1990; Jambon & Johnson, 1997; Slevin, Plant, Lynch, Drinkwater & Gregory, 1988; Joyce, 1988; Bender 1996). The implication is that measures of outcome should take into account individual's self-assessments.

Despite this leading opinion, assessment among children is often conducted using parental reports and the children's satisfaction levels are often ignored. The rationale for asking parents to respond to health-related quality of life issues for their children

is often that parents make the decisions to seek care, accept and conduct the treatment. In addition, instruments for child report are more difficult to develop, because one has to take into account their developmental levels and limitations in comprehension and reading abilities. Doubts about children's capacities to present accurate information, together with easy access to adult sources of information has resulted in an underutilization of self-report assessment data (Stone & Lemanek, 1990).

There are however difficulties with using parental reports. It is known, from health-related quality of life-studies among adult patients and proxy ratings, that ratings provided by others do not correlate highly with ratings provided by patients themselves. Similar results are reported from psychosocial research among children and their parents. Nowadays, the use of reports from different sources, especially in children is recommended (La Greca, 1990a; Achenbach, McConaughy & Howell, 1987).

3.9 THE USE OF MORE THAN ONE INFORMANT

3.9.1 Concurrence between adult patient reports and reports of their proxies

Several studies among adult patients report that proxy ratings are not exchangeable with patient reports. Epstein, Hall, Tognetti & Son (1989), for instance, studied the concurrence between 60 older subjects and their proxies. Subject and proxy ratings were strongly correlated with each other for overall health, functional status, social activity and emotional health and moderately correlated for satisfaction. Mean responses were generally similar for overall health, functional status and social activity. However, those proxies who spent more time per week helping the subject rated the functional status and social activity as more impaired than did the subjects. Subjects who had poorer overall health tended to rate their health relatively lower than did proxies. Rothman, Hedrick, Bulcroft, Hickam and Rubenstein (1991) concluded from a study among patients in an adult day health care center that patient-generated and proxy-generated physical scores were highly correlated, although proxies rated the patients as more impaired than the patients rated themselves. The correlation between psychosocial scores was however not high enough to consider proxy responses as valid substitutes for patient responses. Proxy-generated psychosocial scores were primarily explained by the caregiver's psychological distress and perceived burden. A review of Sprangers and Aaronson (1992) revealed that health care providers and significant others tend to underestimate patient's health-related quality of life and appear to evaluate the patient's health-related quality of life with a similar degree of (in)accuracy.

According to the view that functional status and social activity are readily available for external observation, and emotional status and satisfaction are more likely to be

experienced as private feelings and opinions, it can be concluded that proxies are less accurate than the subjects for the more subjective variables. Proxy ratings appear to be more accurate when the information sought is concrete and observable. Significant others ratings tend to be more accurate when they live in close proximity to the patient, although they can also be biased by the care-giving function of the rater.

3.9.2 Concurrence between child report and parent report

Similar findings were revealed in child and parent reports in pediatrics. Parent and child ratings of nausea in children receiving chemotherapy revealed that their scores correlated significantly. Children, however, generally rated their nausea and emesis as more frequent and more severe than did their parents (Tyc et al., 1993). Research on pain ratings of pediatric cancer patients, revealed also significant correlations between child, parent and nurse ratings, but the correlations were not perfect. Nurses' ratings primarily reflected overt behavioral distress, whereas parents' ratings primarily reflected their own anxiety and their perspective of how much pain their child was experiencing. The child's ratings were primarily associated with child's age, younger children reported less pain (Manne, Jacobson & Red, 1992).

Fritz and Overholser (1989) conducted a study among children with asthma and their parents. To rate the estimated frequency of asthma symptoms, parents were asked to rate the items as they believed the children experienced them. In general, items focusing on physical symptoms tended to obtain higher correlations than items assessing the emotional distress associated with an asthma attack. The highest agreement correlations were on the general physical symptoms factor. Boys showed reasonable agreement with their parents on the panic fear factor whereas females showed no agreement. Asthma symptom factor scores obtained from parents were related to several measures of asthma morbidity, whereas factor scores obtained from children were not. Although it might be concluded that child reports are therefore less valid, a bias may exist as morbidity factors are often also reported by parents.

In a meta-analysis on 119 studies on psychosocial behavior problems, Achenbach, McConaughy and Howell (1987) found a correlation of .25 between parents' and children's scores on behavioral and emotional problems with a higher correlation between 6-11 year olds and undercontrolled problems. The authors concluded that the mean correlation between child report and informant report indicates that proxy reports cannot substitute for self-reports. As in adults, this appears to be most true when the concern is a child's affective experience such as anxiety (Flanery, 1990).

Until more conclusive evidence is obtained indicating that one informant is more reliable, information should be collected from multiple informants especially where children are the focus (Rosenbaum, Cadman & Kirpani, 1990; Eisen, Ware & Brook, 1979; Achenbach, McConaughy & Howell, 1987; La Greca, 1990a). Direct patient feedback can provide valuable information about the significance and impact of

symptoms on daily functioning, whereas information provided by family members is a useful supplement. In studies involving children, parents may serve as a primary data source when necessary, for instance when children are too young, mentally disabled or too sick. However data derived in this indirect manner should be interpreted with great caution (Aaronson, 1990).

As stated above, parental reports on the child's internal states or feelings are less reliable, as these reports consider individual experiences of which parents may not be aware. To obtain a differentiated view of problems in functioning and of the importance of arrays of functioning, children, parents and others may be asked about their own internal states. If children as well as their parents are asked to report on children's health-related quality of life, the question then arises as to whether children are able to report on their own health-related quality of life.

3.10 ARE CHILDREN ABLE TO REPORT ON THEIR HEALTH-RELATED QUALITY OF LIFE?

Although there is certainly a limit as what can be ascertained first hand from children, particularly from young children, this does not mean nothing can be done (Starfield, 1987). Major developmental trends relevant for the assessment among children are described in a review by Stone and Lemanek (1990) in three areas of social and cognitive functioning: the concept of self, understanding of emotions, and language development.

Most self-reports from children are obtained from the third grade reading level (Stone & Lemanek, 1990). Research revealed that children from 7-11 years have self-descriptions that incorporate psychological characteristics and social comparisons. They have an awareness of different components of self, which is situationally based and a differentiation between mental and physical aspects of self. Seven or eight year-old children begin to adjust their self-perceptions according to feedback and past experiences with success or failure. From the age of eight years, children's self-reports become considerable more meaningful as they are better able to report on their thoughts and feelings and to provide more accurate information regarding diverse experiences and situations. The increased use of social comparison processes and the greater psychological awareness of elementary-aged children may, however, contribute to a tendency toward socially appropriate responding (Stone & Lemanek, 1990).

Children in this age group have an accurate recognition of negative emotions, they use mental cues to understand their own emotions and have an understanding of simultaneous experience of different emotions. Whilst six years olds are more apt to use idiosyncratic body cues in identifying their own emotions, children of eight years

and older rely more on their inner experiences and mental cues (Stone & Lemanek, 1990).

The children's language development follows a progression from a concrete action-oriented focus to a more flexible, complex and conceptual understanding and use (Stone & Lemanek, 1990). Pictures can be used to generate interest in the task and to clarify and concretize the verbal material. Another method for engaging the attention of young children is the use of specific relevant age-appropriate questions (Stone & Lemanek, 1990; Flanery, 1990). In addition, the complexities of the test procedures need to match the child's abilities and the questions cannot tap information that exceeds the child's memory capacity. (Flanery, 1990). Studies suggest that children as young as 6-9 year olds are quite reliable reporters about behaviors that require them to use qualitative recall (e.g. type of exercise), but have difficulty with reporting behaviors involving abstract concepts such as time although children as young as 6 years old can be trained in reporting behaviors that require them to use an abstract concept like time (Johnson, Feund, Silverstein, Hansen & Malone, 1990). Even children as young as 5 years old can understand and utilize a numerical rating scale to make a quantitative assessment and extent of bother in a vignette paradigm (Zeltzer, Richie, LeBaron & Reed, 1988). Studies of cognitive development routinely obtain responses to much more complex questions than those required for health assessments from children who are well into the age of concrete operational thought, i.e. by age eight at the latest (Eisen, 1979). The assessment must be sensitive to developmental nuances, not only in understanding, but also in content (Johnson, 1991). Physical, intellectual and emotional functions are constantly changing with normal development. Problems arise when measures developed in an adult population are used among children, as such measures may be misunderstood by children (Johnson, 1991) and items may be irrelevant for children.

3.11 CRITERIA FOR CHILDHOOD HEALTH-RELATED QUALITY OF LIFE MEASURES

Since there is much more variability in health status in children than there is in adults and dysfunctional states show more variability in their manifestation, deviation from the norm is more difficult to detect in children. In addition, we are less sure about society's demands on children and even when we could specify them we are not sure what they are from the children's viewpoint (Starfield, 1987). When looking for evidence of ill health it is not enough to look for deterioration in the curve, but decelerations in the normal rate of increase in the curve must also be watched carefully. Repeated measures are therefore important in order to evaluate progress. In children, it becomes difficult to determine whether a child's failure to achieve independent functioning is part of the normal developmental process, a result of an environment that fosters dependency or loss of ability secondary to illness (Pal, 1996).

Consequently, healthy children have to be included in the reference group when developing generic items and it is advisable to limit the age group to be able to develop a coherent set of relevant items for children. Many instruments have separate questions for those under 5 and for the 6-13 year age group (Pal, 1996).

The content of items, the wording and the layout of the questionnaire have to be adapted for the age group. This means that for primary school children, items have to be relevant, concrete and have to be related to a relatively short period, such as today or this week. Furthermore, pictures that support the meaning of the items can be used to make the questionnaire more attractive.

From the review described above, the following key characteristics of health-related quality of life assessment among primary school children with a chronic illness can be identified as important: (a) the measure should be multidimensional covering the physical, social and psychological consequences of chronic illness and report a profile of the functioning of the child; (b) the instrument has to measure the functioning of the child as well as the evaluation of consequences; (c) the instrument should consist of a generic as well as a disease-specific part. The reference group should include healthy children as well as children with a chronic illness. As the target group of this doctoral dissertation are children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis, items specific for these children have to be included; (d) the measure should include the children's views as well as their parents' on similar subjects so that the measures capture both views, personal evaluations are however only obtainable from self-report; (e) the measure should assess positive as well as negative aspects of health-related quality of life and (f) the instrument should be child-friendly in how it is written and in the concepts used as well as in the lay out.

3.12 WHICH INSTRUMENTS ARE AVAILABLE FOR CHILDREN?

In the following paragraphs characteristics of instruments that have been utilized to assess health-related quality of life issues among primary school children are discussed. In Table 3.1 in Appendix 1 the instruments and their characteristics are presented. Only instruments for healthy children, instruments for the whole group of children with a chronic illness, and children with asthma, diabetes mellitus, epilepsy or juvenile chronic arthritis are included. In addition, only instruments published in Dutch or English are included. Although it must be noted that this is not an exhaustive review, it illustrates which health-related quality of life instruments are currently available for primary school children and what characteristics they have. Since health-related quality of life is a relatively new area of research, nearly all instruments were published relatively recently and used in only one or a few studies. It is therefore difficult to assess the instruments on their

merits. A discussion on whether they meet the characteristics described above is however feasible.

3.12.1 Multidimensionality

As is recommended most instruments cover diverse dimensions and are in this sense multidimensional. For instance, a generic instrument like the Child Health Questionnaire (CHQ) (Landgraf, 1996) provides a profile of 14 health concepts covering the physical domain, the cognitive domain, the social domain and psychological health. A disease-specific instrument like the Paediatric Asthma Quality of Life Questionnaire (Juniper et al., 1996a) covers asthma-specific activity limitations, symptoms, emotional functions and items that children have identified as troublesome. Not all instruments cover however the physical, cognitive, social as well as the emotional domain. In all instruments the physical domain is covered. In two arthritis-specific instruments the physical domain is even the only domain measured (Doherty, Yanni, Conroy & Breshnihan, 1993; Wright et al., 1996). The cognitive domain is missed relatively often (Eisen, 1979; Lewis, Pantell & Kieckhefer, 1989; Collier & MacKinlay, 1997; Juniper et al., 1996a; French & Christie, 1994; Creer, Marion & Creer, 1983; Creer et al., 1993; Doherty, Yanni, Conroy & Breshnihan, 1993; Duffy, Arsenault, Duffy, Paquin & Strawczynski, 1997; Wright et al., 1996).

Most instruments offer a profile, sometimes combined with the possibility of a total score as in the Paediatric Asthma Quality of Life Questionnaire (Juniper et al., 1996a) or the Modified Quality of Life Measure for Youths (Ingersoll & Marrero, 1991). Other instruments, like the FSII-R (Lewis, Pantell & Kieckhefer, 1989; Stein & Jessop, 1990), the Health Resources Inventory (Gesten, 1976), the General Quality of Life Questionnaire (Collier & MacKinlay, 1997), the RAHC measure of Function (Dossetor, Kiddle & Mellis, 1996), and the Pediatric Symptom Checklist (Jellinek et al., 1988) offer only a total sum score and not a profile.

3.12.2 Functioning as well as weighting of importance

Except for a few exceptions, instruments use no evaluations of importance. The following instruments include evaluations in very diverse ways. In the General Child Quality of Life Measure (GCQ), children are asked to respond how they are and then how they would most likely to be (Collier & MacKinlay, 1997). In the TNO-AZL Quality of Life questionnaire (TACQOL), children are asked whether they experience health status problems and their emotional reaction to that problem (Verrips et al., 1997; Vogels et al., 1998). The most extensive exploration of feelings is made by the Child Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997) in which children and parents are asked how well a child functions in a certain area, how upset the child (or parent) is and how satisfied the child (or parent) is. The authors add however that the ratings of functioning, upset and satisfaction might be regarded as redundant and need further research. As recommended in paragraph 3.9.2, the Child Quality of Life

Questionnaire asks the parents not for their child's but their own levels of upset and satisfaction.

Among the chronic illness questionnaires, the Impact of Illness Scale (Hoare & Russell, 1995) for children with epilepsy asks for occurrence as well as the importance of items. In the Childhood Asthma Questionnaire (French & Christie, 1994) children are asked how they function and how they feel when doing certain activities. In the Paediatric Asthma Quality of Life Questionnaire (Juniper et al., 1996a), children are asked to respond to items that the norm group has identified as troublesome. In addition, three items in the activity domain are individualized. This does not reveal however an individual evaluation of consequences as our definition of health-related quality of life implies. In the Juvenile Arthritis Functional Status Index (Wright et al., 1996), children are interviewed about their quality of performance on activities and about which improvement priorities they have.

3.12.3 Measuring generic as well as disease-specific consequences

The Childhood Asthma Questionnaire (French & Christie, 1994) is the only instrument that includes a generic as well as a disease-specific part. This instrument was however only developed and tested for children with asthma. No instruments are available assessing generic as well as chronic illness and disease-specific consequences.

3.12.4 Norm population

All generic instruments are suitable for use among healthy children as well as children with a chronic illness. Most instruments have, however, only recently been developed so that research on other populations than a healthy population has still to be executed. Other instruments like the RAHC (Dossetor, Liddle & Mellis, 1996) and the Paediatric Symptom Checklist (Jellinek et al., 1988) are only suitable for children with a chronic illness.

With regard to disease-specific instruments, most instruments were found for children with asthma. Only one instrument was found for children with diabetes mellitus (Ingersoll & Marrero, 1991). The choice for children with juvenile chronic arthritis was also very limited as two of the three instruments were only directed towards physical functioning.

3.12.5 Child versus proxy report

Several instruments, such as the Child Health Questionnaire (Landgraf, Abetz & Ware, 1996), the TNO-AZL Quality of Life questionnaire (Verrips et al., 1998), the Child Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997), the Asthma Symptom Checklist; the Asthma Problem Behavior Checklist (Creer, Marion & Creer, 1983), the Childhood Assessment Questionnaire (Doherty, Yanni, Conroy & Breshnihan, 1993), the Juvenile Arthritis Quality of Life Questionnaire (Duffy,

Arsenault, Duffy, Paquin & Strawczynski, 1997) have parallel parent and child forms. Whereas most instruments ask the parents about their child's functioning or emotions, the Child Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997) asks for the parents' own feelings of upset and satisfaction. The Paediatric Asthma Quality of Life questionnaire (Juniper et al., 1996a) is accompanied by the Paediatric Asthma Caregiver's Quality of Life Questionnaire (Juniper et al., 1996b). Although using both the child and parent version of this instrument does not provide different observations related to the same subject, it does inform us about the quality of life of other family members than the child itself. This is also of importance as the impact of illness is important for all family members and families' quality of life has a reciprocal effect on the child.

3.12.6 Positive dimensions of health-related quality of life

The TNO-AZL Quality of Life questionnaire is the only instrument with a positive mood dimension (Verrips et al., 1997; Vogels, et al., 1998). The Child Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997) includes a satisfaction scale, but "there is less emphasis on positive aspects of functioning than is usually recommended" according to the authors (p.664).

3.12.7 Suitable for children

Most of the instruments are developed for a relatively large age range, including children from the age of 5-6 years old up to adolescence. This means that the wording as well as the content of items has to be suitable for this large age group. Disease-specific questionnaires are, in particular, developed for large age groups, such as the Paediatric Asthma Quality of Life Questionnaire (Juniper et al., 1996a) and the Quality of Life Scale for children with epilepsy (Wildrick, Parker & Morales, 1996) which are suitable for primary school children up to late adolescence.

The Childhood Asthma Questionnaire form B for 8-11 year olds (French & Christie, 1994), the Child Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997), the Pediatric Symptom Checklist (Jellinek et al., 1988) are examples of questionnaires suitable for a relative coherent age range. The Childhood Asthma Questionnaire has other forms suitable for the age groups of 4-7 and 12-15 year olds (French & Christie, 1994).

The Generic Child Quality of Life Measure (Collier & MacKinlay, 1997) has to be answered by computer and shows pictures which are child-friendly. The Childhood Asthma Questionnaire (French & Christie, 1994) asks children whether they do certain activities and, by means of a "five smiley faces scale" from very happy to very sad, how satisfied they are with these activities. Except for the Juvenile Arthritis Functional Status Index (Duffy, Arsenault, Watanabe, Duffy, Paquin & Strawczynski, 1997) instruments only take a short time to answer by either parents or children.

3.13 CONCLUSION

In this chapter, the definition of health-related quality of life and characteristics of measuring health-related quality of life among children with a chronic illness were delineated. In our opinion, health-related quality of life in children with a chronic illness refers to the functioning of children in the physical, cognitive, social and emotional domains of life, and the gap between their achievements and expectations in their functioning.

Health-related quality of life is a new research area in pediatric health psychology and new instruments have been developed mainly in the past five to ten years so that there are just one or a few studies in which they were used. The experience with health-related quality of life assessment among children is thus very limited. A review of current health-related quality of life instruments revealed that all instruments have merits that have to be checked when looking for a specific measure and all have some apparent relationship with the definition of health-related quality of life. It appeared however that none of the existing instruments fulfilled all the criteria formulated.

We therefore decided to develop the HOW ARE YOU? (HAY), a health-related quality of life questionnaire for children, aged 7-13 years old. The HAY includes a child version and a parent version and consists of a generic section as well as a chronic illness and a disease-specific section. As cognitive and social consequences are relevant for all children, these consequences are included in the generic section. Part of the physical consequences is also generic, such as the ability to participate in daily physical activities. We also decided to include disease-specific physical complaints and selfmanagement issues. As there are many instruments assessing general emotional functioning, it was decided to include illness-related emotions in the HAY. In the next chapter, the developmental stage and the results of a pilot study on the use of the HAY are described.

CHAPTER 4: THE DEVELOPMENT OF THE HOW ARE YOU? (HAY), RESULTS OF THE PILOT STUDY

4.1 INTRODUCTION

In this chapter, the development of the HOW ARE YOU? (HAY), a quality of life questionnaire for children aged 7-13 years old with a chronic illness is described.

The development of the HAY started in 1993 and consisted of three successive stages, including a developmental stage, a pilot study and a main study. During the developmental stage, the definition of quality of life, the construction of an item pool and the choice of response categories were central issues. In the pilot study, the utility, validity and reliability of the preliminary version of the HAY were examined on the basis of which amendments in the HAY were made.

4.2 VALIDITY AND RELIABILITY TESTING

Validity and reliability testing are related to the purposes of the instrument. According to Jaeschke & Guyatt (1990), one can differentiate between at least three basic purposes of quality of life assessment: a) to predict outcome or prognosis, b) to discriminate among individuals along a continuum of health, illness or disability, and c) to evaluate within-person change over time.

The prediction of outcome is related to criterion-validity assumptions, for instance the prediction made from intelligence to school achievement. As is described in Chapter 1 and 2, there are however no direct linear relationships between illness and quality of life. In addition, prediction requires agreement on what the criterion is going to be in a given setting for given individuals. As described in Chapter 3 there were, at the time the research started, no gold standards available for children to assess health-related quality of life. Content validity and construct validity testing were therefore important issues to consider when testing the validity of the HAY.

Content validation is part of construct validation in which the construct to be examined is defined. Construct validity of an outcome measure concerns the theoretical attributes of what is to be measured (Judd & Kenny, 1981) and is assessed under three headings: (a) logical analysis; (b) internal-structure analysis; (c) and cross-structure analysis (Pedhazur & Pedhazur, 1991).

The first strategy for assessing construct validity includes *logical analysis*, which includes the definition of the construct. In addition, item content should be established to perform the dimensions of interest. Items need to be relevant to a large group of subjects in order to discriminate among children (Jaeschke & Guyatt, 1990). Interdisciplinary discussion is therefore required in quality of life research as well as discussion with children and parents to establish constructs and items that are truly useful. Validation among all groups to be assessed is necessary to identify relevant indicators of the constructs (Pal, 1996).

The second strategy for assessing construct validity, *internal-structure analysis*, is most usefully studied through factor analysis, in which observed variables are thought to represent underlying factors that are assumed to be smaller in number than the number of observed variables. It is used widely in psychology and has implications for both the items in a valid and reliable test and for our understanding of the nature of quality of life, but is seldom applied in quality of life research (Welch, 1994).

As health-related quality of life is a subjective construct, referring to the gap between expectations and achievements, items referring to these expectations, preferences or attitudes should therefore be the basis of health-related quality of life assessment. Application of these items as a basis for factor analysis results in a structure that is based on the subjective importance of issues as indicated by the individuals themselves.

When using factor analysis, a distinction must be made among exploratory factor analysis and confirmatory factor analysis. Exploratory factor analysis (EFA) is concerned with the question of how many factors are necessary to explain the relations among a set of indicators. Confirmatory factor analysis (CFA) is concerned with parameter estimation and tests of hypotheses regarding, for example, the number of factors, the indicators belonging to the factors and the relation between the factors. As the theoretical basis of the HAY was identified beforehand, confirmatory factor analysis was used to determine the dimensions.

The third strategy for assessing construct validity, *cross-structure analysis*, refers to tests of hypotheses in which the construct in question is one of the variables (Pedhazur & Pedhazur, 1991). Hypotheses with regard to the relations between the constructs of the new instrument and other validated instruments have to be assessed. Furthermore, hypotheses with regard to differences between distinct groups are used to test construct validity.

These three strategies were used to test the validity of the HAY. In addition, reliability was tested. Reliability refers to the degree to which test scores are free from random errors of measurement. Two indices of reliability can be obtained: test-retest reliability and internal consistency estimates. Ideally, although dependent on

the use of the test, information on both indices should be available (Welch, 1994). Test-retest reliability should be tested by the completion of the HAY at two measurement points in a relatively short period of time, for instance two weeks. As the illness condition of the children may fluctuate in children with chronic illnesses like asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis, it is however possible that a true change in health occurs in just a few days or weeks. Low test-retest reliabilities are in that case difficult to interpret, because it may reflect low reliability, but also true change in the children's condition. Internal consistency estimates, i.e. Cronbach's alphas, are therefore chosen to determine the reliability of the HAY. Internal consistency estimates are based on the notion that, to be substantively meaningful, a composite score has to be based on items measuring the same phenomenon (Pedhazur & Pedhazur, 1991). Internal consistency is a measure of the average correlation among all items in a given subscale, and should be high when the test is reliable.

4.3 RESEARCH QUESTIONS AND HYPOTHESES

The aim of the developmental stage was to formulate the definition of quality of life for children, to formulate items to assess this construct and identify answer categories. These steps belong to the logical analysis. The aim of the pilot study was concerned with internal-structure analysis and cross-structure analysis to obtain preliminary data on the utility, validity and reliability of the pilot version of the HAY.

In addition, as it was theorized that a child version as well as a parent version would be valuable in obtaining information on health related quality of life in this age group on the same issues, the concurrence between parents and children was examined.

The research questions for the pilot study were the following:

- 1. Are children able to answer the HAY within a reasonable period of time?*
- 2. According to the comments of children and parents, which amendments have to be made in the pilot version of the HAY?*
- 3. What is the validity and reliability of the HAY?*
- 4. What is the concurrence between children and parents?*

During the developmental stage, quality of life was defined and relevant indicators of quality of life were composed. A preliminary version of the HAY was constructed.

During the pilot study, internal-structure analysis was performed using confirmatory factor analysis. Hypotheses were formulated relating to the cross-structure analyses (Maes & Bruil, 1995). The first hypothesis concerns the correlation with other measures. The parents also completed the validated Dutch version of the Child

Behavior Check List (CBCL) (Achenbach & Edelbrock, 1983; Verhulst, Koot, & Akkerhuis, 1990). Statistically significant correlations were expected between parts of the CBCL and parts of the parent version of the HAY which measure comparable constructs. As the CBCL is no gold standard, correlations between parts of the CBCL and the HAY were expected to be moderate.

The second hypothesis concerns differences between the total group of children with a chronic illness and healthy children. Although most studies on which the hypotheses are based used parent reports to assess the functioning of children, the following hypotheses are formulated to test differences between groups using parent report as well as child report.

It was hypothesized that children with a chronic illness would score lower on physical, social and psychological functioning (implying a lower prevalence and lower quality of performance levels of relevant behaviors and a higher prevalence of complaints) than healthy children according to child as well as parent report.

The third hypothesis concerns differences between each of the diagnostic groups and the healthy group. Hypotheses were formulated, based on the review of clinical studies among children with asthma, diabetes mellitus, epilepsy, and juvenile chronic arthritis, concerning differences between these four groups and the healthy group.

It was hypothesized that *children with asthma* would experience more physical complaints and more limitations in physical functioning as well as in social functioning compared to healthy children. In addition, absenteeism at school was hypothesized to be higher than among healthy children, implying a lower prevalence of cognitive tasks. It was hypothesized that *children with diabetes mellitus* would have more problems in social functioning. It was hypothesized that *children with epilepsy* would experience more limitations on cognitive and social functioning than healthy children function. Finally, it was hypothesized that *children with juvenile chronic arthritis* would experience more physical complaints and more limitations in physical and social functioning than healthy children. Furthermore, absenteeism at school was hypothesized to be higher than among healthy children, implying a lower prevalence of cognitive tasks.

In addition, differences between each of the disease-specific groups compared to the other disease-specific groups were assessed exploratively. As no clinical studies were found on feelings in general or emotional problems related to illness, no hypotheses were formulated on these issues and assessment would also be explorative.

4.4 SAMPLE

Subjects in the pilot study were 89 children with a chronic illness (30 with asthma, 18 with diabetes mellitus, 22 with epilepsy and 19 with juvenile chronic arthritis) and 134 children who were healthy, aged 8-13 years old. Children who participated in the study had been diagnosed with their illness for three or more months. Other criteria for selection were that they had made a visit to the pediatrician or general practitioner at least once in the last year, that they had no more than one chronic illness and that they were not hospitalized when the questionnaire was administered. In addition, children had to attend normal primary schools. Furthermore, an important requirement was that all children and parents were able to read Dutch.

4.5 PROCEDURE

The parents were contacted by the child's general practitioner (GP), pediatrician or by a patient association. Children with a chronic illness were recruited through their parents. The reference group of healthy children and their parents were recruited through two schools in the west of the Netherlands. Written informed consent was obtained from the parents.

The children with asthma were recruited through GPs¹ (le Coq, 1998). Children with diabetes mellitus, epilepsy and juvenile chronic arthritis were approached by their general practitioner, specialist or patient association. Children with a chronic illness were interviewed at home by a trained interviewer, except for the children with asthma who were interviewed at the medical practice of the GP (le Coq, 1998). The healthy children answered their questionnaire in the presence of an informed schoolteacher; their parents answered the questionnaire at home. Questionnaires were collected by the teacher and returned to the researcher. Children filled in their questionnaire without help from a teacher or trained interviewer although some guidance was given if the child had reading problems. After the questionnaire was completed, children were interviewed about their understanding of the items and the meaning of their answers. In addition, teachers, parents and children were asked to comment on the items and also encouraged to suggest other relevant items. Children were given a small present as incentive fee for their participation; parents were sent a short description of the general outcomes of the pilot study.

¹ The EMGO Institute at Amsterdam who co-operated in the construction of the asthma version of the quality of life questionnaire recruited the group of children with asthma.

4.6 INSTRUMENTS

4.6.1 The developmental stage of the HAY

A study group was formed, involving colleagues within the department of Health Psychology, Leiden University and an external expert on quality of life assessment, in which the structure and content of the questionnaire was discussed. Items were generated from literature as well as semi-structured interviews with eight parents and their children (two interviews per illness group including children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis) and interviews with physicians and patient associations.

Co-operation was started between the EMGO Institute, the VU Amsterdam, and the department of Health Psychology, Leiden University, to develop the asthma-specific part of the questionnaire (le Coq, 1998). The disease-specific part for children with diabetes mellitus was developed in co-operation with a diabetes group that started simultaneously at Rijnland Ziekenhuis, Leiderdorp. The first version of the questionnaire was evaluated by a panel of experts, including two pediatricians and a general practitioner, four psychologists experienced in the assessment of children with a chronic illness and two psychologists experienced in quality of life research. The comments of the panel were incorporated in the pilot version of the "How Are You (HAY)". In accordance with the definition described in Chapter 3 and the areas of importance to children with a chronic illness described in Chapter 2, the HAY was operationalized in the following way.

4.6.2 HOW ARE YOU? (HAY)

The HAY covers the physical, social and psychological consequences of illness for children and consists of a generic, a chronic illness section and a disease-specific section. To make the HAY applicable to primary school children and to be able to measure change over time, items refer to the functioning of the child during the past seven days. In figure 4.1 the structure, dimensions and scales of the HAY are presented graphically.

The *generic section* assesses the child's functioning in daily life areas, areas that are relevant to all children, including healthy children. This section includes six dimensions: 1) physical activities, 2) cognitive tasks, 3) social activities, 4) social problems, 5) general physical complaints, and 6) general treatment characteristics. The physical activities, cognitive tasks and social activities are examined by asking the child about the prevalence of relevant daily activities and tasks, the quality of performance on the activities and tasks and the importance of the activities or tasks (assessed as feelings related to limitations in the quality of performance). Examples of items with regard to the physical activities of children are respectively: (a) "Have you ridden your bike in the past seven days?" (b) "Riding my bike goes...?" and (c) "How do you feel if you have a hard time riding your bike?"

Figure 4.1: Description of the HAY: its sections, dimensions and scales.

			Scales		
			Prevalence ¹	Quality of Performance ²	Feelings ³
HOW ARE YOU? (HAY)	GENERIC SECTION	Physical activities	X	X	X
		Cognitive tasks	X	X	X
		Social activities	X	X	X
		Social problems	X		X
		Physical complaints	X		X
		Treatment			X
	CHRONIC ILLNESS SECTION	Concerns	X		
		Feelings of inferiority	X		
	DISEASE-SPECIFIC SECTION⁴	Physical Complaints	X		X
		Treatment tasks	X		X

1. All prevalence scales include the answer categories yes/no
2. All quality of performance scales include the answer categories very well/well/not so well/not well at all
3. All feelings scales include seven faces ranging from a very happy to a very sad face
4. Disease-specific physical complaints and daily management dimensions are developed for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis

Social problems and physical complaints are both dimensions measured by asking the prevalence of social problems which children may experience at home or at school or general physical complaints as well as the child's related feelings. Examples of items with regard to social problems are respectively: (a) "Have you been teased by other children during the past seven days" and (b) "How do you feel when other children tease you?" Examples of items concerning physical complaints are respectively (a) "Have you had a headache during the past seven days?", and (b) "How do you feel when you have a headache?"

Since data on the prevalence of general treatment characteristics refer to a longer period than the past seven days, they can be more reliably obtained from parents. The

children were only asked about their related feelings. Examples are: "How do you feel if you have to stay in the hospital / go to the doctor?"

The *chronic illness section* consists of items applicable to all children with a chronic illness including two dimensions assessed by prevalence scales: 1) concerns related to having the illness and 2) feelings of inferiority because of having the illness. Examples of items assessing emotions of children with a chronic illness include "Have you felt sad because of your [illness]² during the past seven days?" or "Did you feel you were less important than other children than other children because of your [illness] during the past seven days?"

The third section is developed separately for each disease-specific group. The *disease-specific section* for children with asthma and epilepsy includes a disease-specific complaints dimension, for children with diabetes mellitus and juvenile chronic arthritis both a disease-specific complaints dimension as well as a disease-specific daily management dimension are included.

To examine the most convenient way to assess the different quality of life subjects in children, two versions of the child version of the HAY were developed with a different ordering of items. One version includes items ordered according to their subject, in the other version items are ordered according to their answer category.

A comparable questionnaire for *parents* was developed. The parents were asked comparable questions about the prevalence of consequences and their child's quality of performance on tasks and activities. They were however not asked about their child's feelings but about their own feelings related to the occurrence of problems for their child. The parents thus gave ratings on observable behavior but not on their child's internal processes. This made it possible to evaluate the differential emotional impact of the chronic disease on children and parents.

4.6.3 Child Behavior Checklist (CBCL)

The Child Behavior Checklist (CBCL) was used to test hypotheses with regard to the correlation between matching constructs measured by the CBCL and the HAY. The CBCL (Verhulst, Koot, Akkerhuis & Veerman, 1990) is designed to obtain a systematic report from parents of behaviors observed in children that may reflect psychological difficulties. The measure was standardized within each gender group for children 4-5, 6-11, and 12-16 years of age. Subscales are constructed separately based upon factor analyses for each age and gender group (Verhulst, Koot, Akkerhuis & Veerman, 1990)

² In all emotional items the specific illness the child suffers from is filled in.

4.7 DATA INPUT AND DATA ANALYSIS

The data derived from the HAY were entered by means of the Data entry program of SPSS-PC+ (Norusis, 1988). The data derived from the CBCL were entered by means of the official program diskette. In both programs, ranges were provided for checking valid values of each variable during input. Furthermore, the data were reviewed by using descriptive statistics as Frequencies, Descriptives and Crosstabs.

Internal structure analysis and reliability testing were used to examine the structure and internal consistency of the HAY. Confirmatory factor analysis (CFA) was carried out to test whether the HAY consists of the dimensions as hypothesized using LISREL 7 (Jöreskog & Sörbom, 1989). In addition, the internal consistency of the dimensions was examined using Cronbach's alpha.

CFA was performed on the related feelings items and the disease-specific emotional items of the child-version. The structure found among these items was used to form the related prevalence and quality of performance scales of the child as well as parent-version. As the HAY consists of separate sections, available for different groups and with different answer categories, the LISREL analyses had to be performed separately on the generic section, the chronic illness section and the disease-specific sections. Covariance matrices, assessed by using PRELIS 1.13, were analyzed in LISREL 7 using the maximum likelihood method (ML). The maximum likelihood method was used as it is known that this method is applicable in larger samples but also performs well in small samples (Tanaka, 1987) and is scale invariant (Saris & Stronkhorst, 1984) when items are normally distributed.

Given that it is important not to rely heavily on single measures of fit, the assessment on the fit of the model was based on multiple criteria (Byrne, 1989; Bollen & Long, 1993). Assessment of the overall fit was based on the χ^2/df ratio, the goodness of fit index (GFI), the adjusted goodness of fit index (AGFI), the root-mean-square residual (RMR) and the total coefficient of determination provided by LISREL 7.

A significant chi-square value, relative to the degrees of freedom, indicates that the observed and estimated matrices differ implying that the estimated model does not fit the data. The chi-square estimate is however sensitive to sample size which means that as the sample size increases, the chi-square test has a tendency to indicate a significant probability level which may lead to the rejection of a model which in fact deviates from the population in a trivial way (Schumacker & Lomax, 1996; Tanaka, 1987; Jöreskog & Sörbom, 1989). For well-fitting models the χ^2/df value is smaller than 2.0 (Byrne, 1989). However, ad hoc rules for the retention on well-fitting models on the basis of this statistic have ranged from 2.0 to 5.0 (Bollen & Long, 1993).

The GFI is a measure of how well the variance-covariance matrix from the parameter estimates of the focal model approaches the sample variance-covariance matrix. A GFI or AGFI higher than .80 was considered to be acceptable, a GFI higher than .90 was considered to be moderately good (Jöreskog & Sörbom, 1993), a GFI of .95 or higher was considered to be good. The RMR is a measure of the average of the fitted residuals and can only be interpreted in relation to the size of the observed variances and covariances, the RMR can be used to compare the fit of two different models for the same data (Jöreskog & Sörbom, 1989). Given a good fit of the model, the RMR will be small; this value should be lower than .05 (Byrne, 1989). The total coefficient of determination is a measure of how well the observed variables jointly serve as indicators for all latent factors. A total coefficient of determination of 1.00 is perfect and indicates that the measurement model is perfect (Byrne, 1989; Jöreskog & Sörbom, 1989).

To identify sources of misfit within a specified model, a more detailed evaluation of fit was obtained by inspecting the squared multiple correlations per item (R^2), the standardized residuals, the t-values of the factor loadings and modification indices (MI) as well as the correlations between the latent variables (PHI values) provided by LISREL.

The squared multiple correlation (R^2) is the relative amount of variance in the observed variables which is accounted for by the factors jointly (Byrne, 1989). The absolute size of the residuals is used to verify that for all pairs of variables the “misfit” is low (Bentler & Bonett, 1980). Standardized residuals larger than 2 provide a clue as to a possible mis-specification (Byrne, 1989). The t-values of the factor loadings are defined as the parameter estimate divided by its standard error. Parameters whose t-values are larger than two in magnitude are normally judged to be different from zero (Jöreskog & Sörbom, 1989; Byrne, 1989). The MI are measures associated with the fixed and constrained parameters of the model. For each fixed and constrained parameter the MI is a measure of predicted decrease in chi-square if a single constraint is relaxed and the model is re-estimated (Jöreskog & Sörbom, 1989). The MI were used to inspect whether variables correlate well enough with the corresponding latent variable or with another latent variable. The PHI matrices contain specifications of the correlations between the latent variables. Cronbach's alpha was used to assess the internal consistency of the separate dimensions. When the reliability estimates of distinct dimensions revealed alphas too low (below .70), models were reassessed using LISREL 7.

Normality of scale distributions was inspected by graphical plots. All scales appeared to be approximately normally distributed.

Differences between groups were analyzed by non-paired Student t tests and one-way analyses of variance for continuous data. Pearson chi-square tests of association

were used for nominal data. As sample sizes were small in the group of children with a chronic illness, analyses of variances were explorative and age or gender were not included. A significance level of .01 was used to test differences between the group children with a chronic illness and healthy children. A significance level of .05 was used to test differences between the distinct groups to avoid Type II error because of small sample sizes.

Correlations with other measures were inspected by Pearson correlations. Concurrence between parents and children was established using Pearson correlations and paired Student t tests.

4.8 DEMOGRAPHIC CHARACTERISTICS OF THE SAMPLE

In total 223 children and their parents were included in the study. In Table 4.1 demographic characteristics of the sample are presented.

Table 4.1: Demographic characteristics of the respondents in the pilot study

	Healthy	Asthma	Diabetes mellitus	Epilepsy	Juvenile chronic arthritis
N Boys	72	19	9	8	8
N Girls	62	11	9	14	11
Total number	134	30	18	22	19
Mean Age (SD) (in years)	10.4 (1.3)	9.3 (1.6)	10.8 (1.9)	10.1 (1.7)	10.3 (1.7)
Mean illness duration (SD) (in years)		*	3.66 (2.1)	4.02 (2.6)	3.88 (3.6)

* Data on illness duration were not available for the children with asthma

All healthy children present at school at the time of administration of the questionnaire answered the questionnaire. Children with a chronic illness among this group (n=7) were excluded from analysis. In the healthy group, 12 children were not at school at time of administration and could not answer the questionnaire, 32 (22%) parents of healthy children did not participate. No information is available about the reasons for this or the demographic characteristics of this group. Nor is there any information available about the numbers or characteristics of the non-respondents in the chronically ill group as respondents were recruited by the physicians or patient organizations without keeping records of response percentages.

The mean age of the children with a chronic illness was 10.0 years and the healthy children had a mean age of 10.4 years. There were no statistical differences found in age or gender between both groups. The average age at onset was 7.0 years.

Measurement took place at average 3.8 years after the onset of disease (range 3 months to 9.9 years).

There was no significant difference in gender between the distinct five groups. The mean age of children with diabetes mellitus was however significantly higher than the mean age of healthy children whereas the mean age of children with asthma was significantly lower than the mean age of healthy children ($p < .05$). As group sizes were very small, the representativeness of the groups could not be assessed by comparing these data with prevalence data as described in Chapter 1.

4.9 UTILITY

Almost all of the children were able to answer the questionnaire without experiencing difficulties. A few younger children needed help. In these cases, the interviewer read the questions out loud to the child. The average time children needed to complete the questionnaire were thirty minutes for both versions. The interviews revealed, however, that the children preferred the version with items ordered by their answer categories. The detailed description of how the items had to be answered was not necessary as children understood the system immediately, only the faces scale needed instructions on how to indicate the right face.

The evaluative questions indicated that all items were regarded as relevant, but many children stated that a yes/no-answer to the prevalence questions was too limited. Especially for the items assessing concerns and feelings of inferiority related to having a chronic illness, the impression was that when children experienced these feelings sometimes, but not very often, they gave the answer "no" because this was closer to their answer and more easy to admit than "yes". In addition, it was felt that the negative answer categories of the items assessing quality of performance had more diagnostic relevance, as it was difficult for children to differentiate between well and very well.

The seven faces included in the related feelings items were considered to be too many, as it was difficult for children to differentiate between the three happy faces. In addition, the neutral middle category was seen by many children as a glum face. For some children it was difficult to answer the related feelings items as the emotion they felt was other than happy or sad, for instance one child said that he "was not happy or sad but angry when he was teased". In addition, as a result of the interviews, it was learned that fourteen items had to be reformulated.

Table 4.2: Lambda-values, corrected item-total-correlations and Cronbach's alphas for the generic section of the HOW ARE YOU? (HAY)

Factor I-VI Items	Lambda ^a	Item-total corr. ^b	α^c	Item-total corr. ^d	α^e
I. Physical activities			.81		.90
How do you feel when you have trouble					
biking	.56	.53		.67	
playing outside	.72	.67		.71	
playing a certain sport	.69	.61		.78	
joining in gym	.67	.58		.78	
swimming	.73	.58		.78	
II. Cognitive tasks			.76		.88
How do you feel when you have trouble					
making homework	.47	.45		.74	
paying attention	.63	.55		.76	
doing assignments	.70	.60		.82	
reading at home	.59	.49		.67	
drawing	.65	.55		.51	
III. Social activities			.79		.76
How do you feel when you have trouble					
going out	.59	.50		.55	
playing with friends	.72	.62		.65	
visiting people	.66	.60		.54	
playing with pets	.59	.55		.26	
playing at school	.69	.57		.58	
IV. Social problems			.78		.85
How do you feel when you					
are teased	.64	.55		.70	
are laughed at	.75	.63		.75	
are left out of certain activities	.83	.67		.71	
V. Physical problems			.77		.77
How do you feel when you					
have trouble sleeping	.63	.55		.42	
have a headache	.75	.62		.63	
are tired	.58	.51		.55	
have stomach aches	.74	.61		.68	
VI. Treatment			.70		.75
How do you feel when you					
have to take medicine	.45	.41		.39	
have to have a shot	.61	.48		.49	
have to got to the GP	.80	.58		.65	
have to go to the specialist	.70	.57		.57	
have to stay in the hospital	.27	.27		.41	

^a All lambda-values are significant at $p < .01$

^b Item-total correlations child version

^c Cronbach's alphas child version

^d Item-total correlations parent version

^e Cronbach's alphas parent version

4.10 INTERNAL STRUCTURE AND RELIABILITY OF THE GENERIC SECTION OF THE HAY

Confirmatory factor analysis was carried out using LISREL 7 (Jöreskog & Sörbom, 1989) to determine whether the HAY consisted of dimensions as was theorized. The model assessing the generic section of the HAY, consisting of six dimensions, revealed a chi-square of 538.35 ($N=217$, $Df=309$, $p<.001$). The χ^2/df ration was lower than 2. The goodness-of-fit index (GFI) was .84. The root mean square residual was 0.06. The lambda values, which can be seen as factor loadings, ranged from .27 to .83 and were significant at a level of $p < 0.01$. Two items were deleted because the factor loadings were too low. The alpha coefficients of the generic scales of the child version ranged from .70 to .81. The alpha coefficients of the generic scales of the parent version ranged from .75 to .90 (see next page for Table 4.2 for lambda values, item-total correlations and Cronbach's alphas).

The PHI values ranged from .25 to .85 (see Table 4.3a and 4.3.b for PHI values and Pearson correlations). The high correlation between the first three factors (physical activities, cognitive tasks and social activities) suggested that they could be seen as one factor. To decide whether the latent factors were independent, the Pearson r should be substantially lower than the mean Cronbach's alpha. Examination of the Pearson r and mean Cronbach's alpha of the dimensions revealed that these three factors represented enough unique variance.

Table 4.3a: PHI values between the dimensions of the generic section of the HOW ARE YOU? (HAY)

Factor I-VI		PHI values					
		I	II	III	IV	V	VI
I	Physical activities	1.00					
II	Cognitive tasks	.77	1.00				
III	Social activities	.70	.85	1.00			
IV	Social problems	.45	.52	.66	1.00		
V	Physical complaints	.26	.41	.36	.46	1.00	
VI	General treatment	.25	.28	.27	.30	.51	1.00

Table 4.3b: Pearson correlations between the dimensions of the generic section of the HOW ARE YOU? (HAY)

Factor I-VI		Pearson r					
		I	II	III	IV	V	VI
I	Physical activities	1.00					
II	Cognitive tasks	.58*	1.00				
III	Social activities	.54*	.64*	1.00			
IV	Social problems	.33*	.41*	.52*	1.00		
V	Physical complaints	.23*	.34*	.32*	.38*	1.00	
VI	General treatment	.20*	.20*	.25*	.24*	.42*	1.00

$p < .01$

The structure of the six dimensions, which was based on the related feeling scales, was accepted and used to form the corresponding prevalence and quality of performance scales. As described above, physical activities, cognitive tasks and social activities are examined by asking the child about the prevalence of relevant daily activities and tasks, the quality of performance on the activities and tasks and the importance of the activities or tasks. These dimensions each include thus three scales of 5 items. The social problems and physical problems are examined by two scales asking about the prevalence of problems and the importance of problems and consist each of two scales of respectively 3 and 4 items. The treatment scale is only measured by the importance of treatment activities and includes 5 items. The generic section of the pilot version of the HAY consists thus of 64 items.

4.11 INTERNAL STRUCTURE AND RELIABILITY OF THE CHRONIC ILLNESS AND DISEASE-SPECIFIC SECTIONS

Inspection of the distribution of the answer categories of the subsection measuring feelings of inferiority and concerns related to having a chronic disease revealed that these dichotomous items were very skewed so that further analyses of this subsection were not possible.

As the distinct illness groups had too small a sample size, LISREL analyses were not performed on the subsection measuring disease-specific physical complaints and daily management. The internal consistency of the disease-specific complaints and disease-specific management scales ranged from .69 to .91 (see Table 4.4 to 4.7).

The *asthma-specific complaints section* included only one dimension, including 5 items (Table 4.4) which had a Cronbach's alpha of .80. No items were deleted. This structure was used to form the related prevalence scales, so that the asthma-specific subsection consists of two scales each consisting of 5 items measuring prevalence of complaints and the feelings related to complaints. The asthma-specific section therefore now includes 10 items.

Table 4.4: Internal consistency of the disease-specific sub-section for children with asthma

Dimension/ item	Item-total correlation child version	Alpha child version	Item-total correlation parent version	Alpha parent version
I Physical complaints		.80		.74
How do you feel when you				
- have to cough	.63		.59	
- are short of breath	.59		.65	
- are wheezy	.57		.56	
- having had an asthma seizure	.59		. ¹	
- awake at night	.51		.33	

¹ Due to an error, this item was missing in the parent version

The *diabetes mellitus-specific section* included two dimensions, measuring *complaints* (4 items) and *daily management* (4 items), the Cronbach's alphas were respectively .75 and .89 (Table 4.5).

Table 4.5: Internal consistency of the disease-specific sub-section for children with diabetes mellitus

Dimension/ item	Item-total correlation child version	Alpha child version	Item-total correlation parent version	Alpha parent version
I Physical complaints		.75		.75
How do you feel when you				
- have hypoglycemia	.38		.41	
- have hyperglycemia	.49		.58	
- are dizzy	.61		.70	
- have bruises from insulin injections	.69		.46	
II Daily management		.89		.87
How do you feel when you				
- have to check blood sugar level	.78		.79	
- have to have insulin injections	.70		.63	
- have a HbA1C-shot	.78		.84	
- change the place you inject	.77		.65	

One item of the complaints scale and two items of the daily management scale had to be deleted because of decreasing alpha. This structure was then used to form the related prevalence scales, so that the diabetes-specific section consists of a scale measuring the prevalence of complaints, a related scale measuring feelings towards complaints and a scale measuring the prevalence of daily management together with a related scale measuring feelings towards daily management. The diabetes-specific section now includes therefore 18 items.

Table 4.6: Internal consistency of the disease-specific sub-section for children with epilepsy

Dimension/ item	Item-total correlation child version	Alpha child version	Item-total correlation parent version	Alpha parent version
I Physical complaints		.69		.71
How do you feel when you				
- you are nauseous	.39		.16	
- you fall	.38		.43	
- are dizzy	.54		.64	
- have seizures	.37		.45	
- get up tired	.53		.73	

The *epilepsy-specific complaints section* included only one dimension of 5 items. The Cronbach's alpha of this dimension was .69, no item had to be deleted (Table 4.6). This structure was thus used to form the related prevalence scale, so that the epilepsy-specific section consists of a prevalence scale measuring the prevalence of

physical complaints and a scale measuring the feelings towards these complaints. The epilepsy-specific section now includes therefore 10 items.

Table 4.7: Internal consistency of the disease-specific sub-section for children with juvenile chronic arthritis

Dimension/ Item	Item-total- correlation child version	Alpha child version	Item-total- correlation parent version	Alpha parent version
I Physical complaints		.91		.88
How do you feel when you				
- have trouble moving	.90		.86	
- have swollen joints	.72		.67	
- have painful joints	.84		.79	
II Daily management		.82		.79
How do you feel when you				
- have to do exercises	.57		.75	
- have to have physical therapy	.68		.72	
- have to wear (one or more) splints	.62		.62	
- have to take your arthritis into account	.78		.27	

The juvenile chronic arthritis-specific section included two dimensions, measuring complaints (3 items) and daily management (4 items). The Cronbach's alphas of these dimensions were respectively .91 and .82 (Table 4.7).

Two items of the complaints scale and one item of the management scale had to be deleted because of decreasing alpha. This structure was then used to form the related prevalence scales, so that the arthritis-specific sub-section consists of two prevalence scales measuring respectively the prevalence of physical complaints and daily management and two related scales measuring the feelings towards respectively the physical complaints and daily management. The arthritis-specific section therefore now includes 14 items.

4.12 CORRELATIONS BETWEEN SUBSCALES OF THE HAY-PARENT VERSION AND CBCL

It was hypothesized that the scales of the CBCL assessing “physical, cognitive and social competence, social problems and physical complaints” would measure constructs comparable to the scales of the HAY measuring “quality of performance on respectively the physical activities, cognitive tasks, social activities, and the prevalence of social problems and physical complaints”. Pearson correlations between the scales of both instruments were computed. In Table 4.8 the correlations between the scales are presented. The correlations ranged from .30 to .51 and were statistically significant at a $p < .01$ level. All correlations were moderate.

Table 4.8: Pearson correlations between scales of the CBCL and scales of the parent version of the HAY

CBCL	HAY parent version	Pearson R
Activity competence	Quality of performance on physical activities	.30*
School competence	Quality of performance on cognitive activities	.46*
Social competence	Quality of performance on social activities	.39*
Social problems	Social problems	-.51*
Physical complaints	Physical complaints	-.48*

*p<.01

4.13 CHILDREN WITH A CHRONIC ILLNESS VERSUS HEALTHY CHILDREN: CHILD REPORT

Another important issue is whether the HAY differentiates between children with a chronic illness and healthy children. Items were recoded so that a high score indicates a high quality of life, meaning a good level of functioning, a good quality of performance or less negative feelings.

Table 4.9: Means and standard deviations of children with a chronic illness and healthy children: child report

Scales	CHILD REPORT	
	Ill M (SD)	Healthy M (SD)
Prevalence		
Physical activities	1.64 (.23)**	1.75 (.18)
Cognitive tasks	1.68 (.27)**	1.80 (.17)
Social activities	1.69 (.21)#	1.75 (.18)
Social problems	1.80 (.30)	1.81 (.27)
Physical complaints	1.60 (.30)*	1.71 (.28)
Quality of performance		
Physical activities	3.20 (.57)**	3.53 (.37)
Cognitive tasks	3.20 (.51)	3.30 (.41)
Social activities	3.32 (.48)#	3.45 (.41)
Feelings		
Physical activities	3.73 (1.20)#	3.35 (1.24)
Cognitive tasks	3.52 (1.07)	3.38 (1.05)
Social activities	3.12 (1.22)	3.00 (1.02)
Social problems	2.42 (1.05)	2.46 (1.11)
Physical complaints	2.90 (.99)	3.05 (.92)
Treatment	3.89 (1.04)*	3.52 (1.02)

Note. A high score indicates a high quality of life, meaning a good level of functioning, a good quality of performance or less negative feelings

** p<.001

* p<.01

p<.02

In Table 4.9 the means and standard deviations of both groups are presented as well as the statistically significant differences between both groups. Comparison of children's scores using Student t tests revealed statistically significant differences in the expected direction on the prevalence of physical activities ($p < .001$), on the prevalence of cognitive tasks ($p < .001$), on the prevalence of physical complaints ($p < .01$), and on the quality of performance on physical activities ($p < .001$). In contrast, children with a chronic illness scored, on an average, significantly more positively than healthy children on the feelings scale related to general medical treatment ($p < .01$).

Trends were found on the prevalence of social activities scale on which children with a chronic illness scored lower than healthy children ($p < .02$). In addition, children with a chronic illness scored lower than healthy children on the quality of performance on social activities ($p < .02$). In contrast, children with a chronic illness scored more positive feelings related to limitations in physical activities than healthy children ($p < .02$) did.

Table 4.10: Means and standard deviations of children with a chronic illness and healthy children: parent report

Scales	PARENT REPORT	
	Ill M (SD)	Healthy M (SD)
Prevalence		
Physical activities	1.70 (.23)**	1.85 (.15)
Cognitive tasks	1.68 (.24)**	1.82 (.17)
Social activities	1.74 (.21)*	1.83 (.16)
Social problems	1.78 (.34)	1.86 (.24)
Physical complaints	1.63 (.30)**	1.85 (.23)
Quality of performance		
Physical activities	3.09 (.60)**	3.47 (.38)
Cognitive tasks	2.92 (.55)*	3.11 (.44)
Social activities	3.21 (.45)#	3.35 (.44)
Feelings		
Physical activities	2.27 (.83)*	2.56 (.69)
Cognitive tasks	2.17 (.77)	2.34 (.58)
Social activities	2.11 (.61)	2.29 (.58)
Social problems	1.66 (.72)**	2.00 (.68)
Physical complaints	2.17 (.61)**	2.51 (.54)
Treatment	2.82 (.65)	2.92 (.57)

Note. A high score indicates a high quality of life, meaning a good level of functioning, a good quality of performance or less negative feelings

** $p < .001$

* $p < .01$

$p < .02$

4.14 CHILDREN WITH A CHRONIC ILLNESS VERSUS HEALTHY CHILDREN: PARENT REPORT

In general, it can be concluded that parents of children with a chronic illness scored lower on all scales lower than parents of healthy children (see Table 4.10 for means and standard deviations).

Comparison of the parents' scores using Student t tests revealed that parents of children with a chronic illness scored significantly lower than parents of healthy children on the prevalence of physical activities ($p < .001$), cognitive tasks ($p < .001$), social activities ($p < .01$), and physical complaints ($p < .001$), and on the scales measuring the quality of performance on physical activities ($p < .001$) and cognitive tasks ($p < .01$). In addition, parents of children with a chronic illness scored significantly more negative feelings towards limitations on physical activities ($p < .01$), social problems ($p < .001$) and physical complaints ($p < .001$). Trends were found on the quality of performance on social activities on which parents of children with a chronic illness scored lower than parents of healthy children ($p < .02$) did.

4.15 DIFFERENCES BETWEEN GROUPS

Table 4.11: Mean scores and standard deviations of distinct groups according to child report

	Asthma M (SD)	IDDM M (SD)	Epilepsy M (SD)	JCA M (SD)	Healthy M (SD)
Prevalence					
Physical activities	1.63 (.22)	1.57 (.18) ^h	1.75 (.20)	1.57 (.26) ^h	1.75 (.18)
Cognitive tasks	1.64 (.25) ^h	1.68 (.29)	1.70 (.24)	1.69 (.32)	1.80 (.17)
Social activities	1.65 (.22)	1.76 (.22)	1.73 (.16)	1.64 (.24)	1.75 (.18)
Social problems	1.73 (.34)	1.89 (.23)	1.74 (.29)	1.88 (.27)	1.81 (.27)
Physical complaints	1.55 (.31)	1.64 (.25)	1.68 (.27)	1.55 (.37)	1.71 (.28)
Quality of performance					
Physical activities	3.16 (.64) ^h	3.31 (.56)	3.29 (.50)	3.05 (.57) ^h	3.53 (.37)
Cognitive tasks	3.16 (.58)	3.20 (.34)	3.04 (.56)	3.42 (.40)	3.30 (.41)
Social activities	3.18 (.60)	3.37 (.32)	3.42 (.39)	3.36 (.48)	3.45 (.41)
Feelings					
Physical activities	3.82 (1.22)	3.84 (.78)	3.82 (1.56)	3.39 (1.00)	3.35 (1.24)
Cognitive tasks	3.49 (1.19)	3.58 (.93)	3.66 (1.13)	3.36 (.95)	3.38 (1.05)
Social activities	2.82 (1.20) ^e	3.10 (.71)	3.81 (1.52) ^{ha}	2.80 (.99)	3.00 (1.02)
Social problems	2.17 (.97)	2.63 (.94)	2.59 (1.23)	2.40 (1.02)	2.46 (1.11)
Physical complaints	2.93 (1.18)	3.13 (.64)	2.61 (.95)	2.95 (.95)	3.05 (.92)
Treatment	3.86 (1.05)	4.01 (.87)	4.18 (1.08)	3.49 (1.10)	3.52 (1.02)

^h Mean significant different from healthy
^a Mean significant different from asthma
^d Mean significant different from diabetes mellitus (IDDM)
^e Mean significant different from epilepsy
^j Mean significant different from juvenile chronic arthritis (JCA)

To test several hypotheses related to differences between specific groups of children with a chronic illness and healthy children, one-way-analyses of variance (ANOVA) were carried out (see Table 4.11 and Table 4.12 for means and standard deviations of respectively the child and parents version).

The hypotheses related to *children with asthma* were partially confirmed. As was hypothesized, children with asthma had a significantly lower prevalence of physical activities and more physical complaints according to the parents than healthy children ($p < .05$). In addition, children themselves scored a lower quality of performance on physical activities ($p < .05$). Furthermore, both asthmatic children and their parents reported a statistically significant lower prevalence of cognitive tasks compared to healthy children ($p < .05$). In contrast to what was expected, no significant results on social activities or social problems were found. Compared to the other disease-specific groups, children with asthma had a significantly higher quality of performance on physical activities compared to children with juvenile chronic arthritis according to the parents. In addition, children themselves scored more negative feelings towards limitations in social activities compared to children with epilepsy ($p < .05$).

Table 4.12: Mean scores and standard deviations of distinct groups according to parent report

	Asthma M (SD)	IDDM M (SD)	Epilepsy M (SD)	JCA M (SD)	Healthy M (SD)
Prevalence					
Physical activities	1.72 (.22) ^h	1.68 (.24) ^h	1.80 (.19) ^j	1.57 (.24) ^{he}	1.85 (.15)
Cognitive tasks	1.67 (.27) ^h	1.76 (.20)	1.67 (.22)	1.64 (.26) ^h	1.82 (.17)
Social activities	1.74 (.13)	1.74 (.24) ^j	1.86 (.18)	1.65 (.24) ^{hd}	1.83 (.16)
Social problems	1.73 (.40)	1.88 (.26)	1.70 (.37)	1.86 (.23)	1.86 (.24)
Physical complaints	1.61 (.30) ^h	1.69 (.28)	1.65 (.30)	1.55 (.32) ^h	1.85 (.23)
Quality of performance					
Physical activities	3.24 (.56) ^j	3.32 (.42) ^j	3.16 (.52) ^j	2.54 (.62) ^{ehad}	3.47 (.38)
Cognitive tasks	3.06 (.63)	3.08 (.25)	2.64 (.54) ^h	2.87 (.53)	3.11 (.44)
Social activities	3.30 (.36)	3.28 (.41)	3.21 (.52)	3.00 (.49) ^h	3.35 (.42)
Feelings					
Physical activities	2.21 (.86)	2.72 (.85)	2.23 (.78)	1.97 (.70) ^h	2.56 (.69)
Cognitive tasks	2.17 (.82)	2.47 (.76)	2.04 (.69)	2.04 (.74)	2.34 (.58)
Social activities	2.18 (.78)	2.22 (.45)	1.88 (.55)	2.13 (.49)	2.29 (.58)
Social problems	1.77 (.92)	1.89 (.66)	1.61 (.62)	1.33 (.40) ^h	2.00 (.68)
Physical complaints	2.15 (.62)	2.50 (.57) ^j	2.21 (.64)	1.86 (.48) ^{hd}	2.51 (.54)
Treatment	2.78 (.73)	3.00 (.46)	2.95 (.56)	2.56 (.72)	2.92 (.57)

^h Mean significant different from healthy
^a Mean significant different from asthma
^d Mean significant different from diabetes mellitus (IDDM)
^e Mean significant different from epilepsy
^j Mean significant different from juvenile chronic arthritis (JCA)

In contrast to what was expected, *children with diabetes mellitus* scored significantly lower on the prevalence of physical activities according to child as well as parent report than healthy children ($p < .05$), while no significant differences were found on social activities. Compared to the other disease-specific groups, parents of children with diabetes mellitus scored a higher prevalence of social activities and a better quality of performance on physical activities of their children than parents of children with juvenile chronic arthritis did. In addition, parents of children with diabetes mellitus scored fewer negative feelings towards physical complaints than parents of children with juvenile chronic arthritis did.

As hypothesized, *children with epilepsy* scored a significant lower quality of performance on cognitive tasks than healthy children according to their parents ($p < .05$). No significant results were found for limitations on cognitive tasks in child report or limitations on social activities or social problems in parent or child report. In contrast, children with epilepsy themselves scored significantly higher on feelings related to difficulties in social activities than healthy children, revealing fewer negative feelings. Compared to the other disease-specific groups, children with epilepsy scored higher on the prevalence and quality of performance on physical activities than children with juvenile chronic arthritis according to parent reports ($p < .05$). In addition, children with epilepsy scored significantly higher on feelings related to difficulties in social activities than children with asthma did, revealing fewer negative feelings.

As hypothesized, *children with juvenile chronic arthritis* scored significantly lower on the prevalence and quality of performance on physical activities than healthy children according to child report ($p < .05$). According to their parents' reports, children with juvenile chronic arthritis scored significantly lower on the prevalence of physical activities, lower on the prevalence of cognitive tasks and social activities, lower on the (absence of) physical complaints, lower on the quality of performance on physical activities and lower on the quality of performance on social activities than healthy children ($p < .05$). In addition, parents of children with juvenile chronic arthritis scored significantly lower on the related feelings towards limitations in physical activities and lower on feelings related to social problems and physical complaints than parents of healthy children did, revealing more negative feelings ($p < .05$). Compared to the other disease-specific groups, according to parent reports, children with juvenile chronic arthritis scored significantly lower on the prevalence of physical activities compared to children with epilepsy, lower on the prevalence of social activities than children with diabetes mellitus, and lower on the quality of performance on physical activities than all other groups ($p < .05$). In addition, parents of children with juvenile chronic arthritis scored significantly lower on feelings towards physical complaints than parents of children with diabetes mellitus did ($p < .05$).

4.16 CONCURRENCE BETWEEN THE CHILD AND THE PARENT VERSION

In Table 4.13 the correlations between the scores on the prevalence and quality of performance scales of parents and children are presented. The correlations between the child- and the parent version range from .15 to .57 and are significant on all scales, except for the scale measuring the quality of performance on social activities. The correlation between parents and children was the highest on the prevalence of physical activities and the prevalence of physical complaints. In general, the correlations between parents and children were higher on the prevalence scales than on the quality of performance scales. Concurrence between the feelings scales was not assessed because of different content.

Table 4.13: Pearson correlations between the child and parent version of the HAY

Scales	N	Pearson R
Prevalence		
Physical activities	187	.57*
Cognitive tasks	183	.45*
Social activities	185	.21*
Social problems	183	.39*
Physical complaints	182	.52*
Quality of performance		
Physical activities	185	.24*
Cognitive tasks	181	.34*
Social activities	184	.15

* $p < .01$

In Table 4.11 and 4.12 the means and standard deviations of parent report and child report are presented. Comparison of the mean scores of the parents with the mean scores of their children using paired t tests on the prevalence and quality of performance scales revealed that the parents scored significantly higher on the prevalence of physical activities ($t(186) = -5.23$; $p < .001$), social activities ($t(184) = -3.64$; $p < .001$), and physical complaints ($t(181) = -3.35$; $p < .001$), which means that parents scored a higher prevalence of activities and a lower prevalence of physical complaints. In contrast, the parents scored significantly lower than their children on the quality of performance on cognitive tasks ($t(180) = 5.24$; $p < .001$) and social activities ($t(185) = 2.54$; $p < .01$). The differences in means on the related feelings scales were not assessed because of a different content.

4.17 CONCLUSION AND DISCUSSION

Although revisions of the HAY and further testing of validity and reliability are needed, it can be concluded that the preliminary data on the utility, validity and reliability of the HOW ARE YOU? (HAY) are encouraging.

4.17.1 Utility

All the children except a few younger ones were able to complete the questionnaire independently in an average time of 30 minutes. It was therefore concluded that the HAY is suitable for children of seven years and older. Revisions of the preliminary version of the HAY appeared however to be necessary on the basis of comments of parents and children and results of analyses.

Many children stated that a yes/no-answer to the prevalence questions was too limited. In particular, for the items assessing emotions related to having a chronic illness, the impression was that children needed more answer categories to express their meaning. It was therefore decided to insert four answer categories from “no, never” to “yes, very often” to the prevalence items.

In addition, the answer categories for the quality of performance scales and feelings scales were reformulated into one positive and three negative categories because the negative scorings had more diagnostic relevance. As some children had difficulty with answering the related feelings scales because they felt another emotion than happy or sad, the related feelings items had to be reformulated into items assessing a more cognitive weighting of bother.

Two dimensions measuring happy feelings and negative feelings had to be inserted as it was felt that the happy feelings were important to include (Hyland, 1992, Nordenfelt, 1995) and the negative feelings only were assessed in relation to chronic illness. Finally, as the format with items ordered according to their answer categories, and not item content, was more convenient for children, this format was chosen.

4.17.2 Confirmatory factor analysis and reliability testing

Confirmatory factor analysis revealed an acceptable fit for the model of six dimensions in the generic section. On inspecting the interpretability of the model (Browne & Cudeck, 1992) and the good internal consistency of all six dimensions, it was decided to accept the model temporarily for re-examination in the main study on a large sample of children, with a revised version of the items and answer categories.

The internal structure of the chronic illness and disease-specific sections section could not be determined because of high skewedness of answer categories of items measuring emotions related to having a chronic illness and sample sizes that were too small for the dimensions measuring disease-specific complaints and daily management characteristics. Reliabilities of the disease-specific dimensions were ranging from acceptable to good, only the disease-specific complaints dimension for children with epilepsy was considered to be too low.

4.17.3 Concurrence with the CBCL

The correlations between subscales of the parent-version of the HAY with corresponding scales of the CBCL were moderate. The correlations between the scales measuring cognitive competence, prevalence of social problems and physical complaints of both instruments were higher than the correlations between the quality of performance on physical and social activities scales of the HAY and physical and social competence scales of the CBCL. An explanation for this discrepancy may be that the physical competence and social competence scales of the CBCL measure not only quality of performance but also the involvement of children in physical and social activities, whereas the quality of performance scales of the HAY measure only competence. The correlations between the CBCL and HAY support the validity of the HAY as high correlations cannot be expected when there are no gold standards (Hillers, 1994).

4.17.4 Differences between groups

As hypothesized, children with a chronic illness scored lower than healthy children according to the children and/or their parents on almost all prevalence or quality of performance scales. This implies that the HAY differentiates between the prevalence and quality of performance concerning physical, cognitive and social functioning of children with a chronic illness and healthy children. It must be noted, however, that a small part of the children with a chronic illness were interviewed just during the beginning of the summer vacation. Results concerning the prevalence of cognitive tasks could be influenced by this. In the main study, assessments should be carried out after the vacation period.

The fact that children with a chronic illness scored more positive feelings related to general treatment characteristics than healthy children should not be surprising. Children with a chronic illness have more experience with treatment and thus may rate the not especially invasive aspects of general treatment, as going to the doctor, as less distressing than healthy children do. A trend was found concerning the children's feelings related to limitations in physical activities, on which children with a chronic illness rated less distressful than healthy children did. Parents of children with a chronic illness reported more negative feelings towards their child's limitations in physical activities, social problems and physical complaints than parents of healthy children did. These results emphasized the relevance of the feeling scales for both children and their parents. As stated in Chapter 3, quality of life is not only a question of (quality of) performance, it is also concerned with the importance of this performance to the individual. The most important issue, however, is the relation between the individual level of functioning and the related feelings.

In contrast to what was expected, no significant differences were found on the prevalence of social problems scale of the child report or parent report, nor on the quality of performance on cognitive tasks scale of the child report. The number of

studies on which the hypotheses on social functioning were based was, however, small. It is not clear whether the pilot version of the HAY fails to detect social problems in children with a chronic illness or whether social problems do not occur more often in children with a chronic illness.

Despite the small sample sizes of the disease-specific groups, results with regard to differences between disease-specific groups and healthy children were encouraging as part of the hypotheses was confirmed.

Children with asthma scored more limitations in the quality of performance on physical activities, whereas according to the parents' reports, they scored a lower prevalence of physical activities. In addition, children with asthma had more general physical complaints than healthy children according to the parents. Furthermore, both children with asthma and their parents reported a lower prevalence of cognitive tasks, which is in accordance with the conclusions stated in Chapter 2, that school absenteeism is relatively high in this group, which may lead to a lower prevalence of cognitive tasks. In contrast to what was expected, children with asthma did not have more limitations in social activities or more social problems than healthy children. The number of clinical studies on which this hypothesis was based was however low. One study even found no differences in social functioning between children with asthma and healthy children (Graetz & Shute, 1995). Only one significant result between disease-specific groups was found in the child reports in which children with asthma scored more negative feelings towards limitations on social activities than children with epilepsy.

Contrary to our expectations, children with diabetes mellitus and their parents scored a lower prevalence of physical activities than healthy children, a difference which is not easy to explain. In addition, in contrast to what was expected, no significant results on social activities or problems were found. As was the case among children with asthma, however, the number of studies on which the hypothesis on social functioning was based was small. In addition, other mediating factors than illness, such as time of onset and gender, were reported to be important in this (Rovet, Ehrlich & Hoppe, 1987).

As hypothesized, parents of children with epilepsy reported a lower quality of performance on children's cognitive tasks than parents of healthy children did. In contrast to what was expected, no significant results were found on social activities or social problems. This hypothesis was, however, also based on just two studies, indicating that children with epilepsy were at risk of developing problems in social functioning (Dorenbaum, Cappelli, Keene & McGrath, 1985; Hoare & Kerley, 1991). Children with epilepsy even scored fewer negative feelings related to limitations in social activities than healthy children or children with asthma, which is not easy to explain.

As hypothesized, children with juvenile chronic arthritis had a lower prevalence of and a lower quality of performance on physical activities than healthy children according to both parents and children. Furthermore, the parents scored also a lower prevalence of cognitive tasks and social activities, a lower quality of performance on social activities and more physical complaints. In addition, the parents scored more negative feelings related to physical activities, social problems, and physical complaints than parents of healthy children did. The higher prevalence of limitations or complaints combined with the negative evaluation of these limitations or complaints, reveal that these issues are important in the counseling of parents with children with juvenile chronic arthritis.

With regard to differences between groups, while taking notice of the disease-specific symptoms, it should not be surprising that, according to their parents, children with juvenile chronic arthritis are more limited in the quality of performance on physical activities than all other groups, although also children with asthma were expected to have limitations in this area. In addition, children with juvenile chronic arthritis have a lower prevalence of physical activities than children with epilepsy and a lower prevalence of social activities than children with diabetes according to their parents. This might be a result of the fact that the social activities dimension also involves physical activities with others in which children with juvenile chronic arthritis are more limited because of their physical limitations. In addition, parents of children with juvenile chronic arthritis scored more negative feelings on physical complaints than parents of children with diabetes mellitus.

The fact that more significant results were found on parent reports, especially for children with juvenile chronic arthritis, is difficult to explain. Whether parents overreport or children underreport is not known. Until the contrary is proven, it is suggested that both versions should be included in assessing quality of life among primary school children.

The reliability and validity of the scales measuring emotions related to having a chronic illness could not be assessed so that groups could not be compared on these scales.

Although not all the hypotheses formulated were confirmed, the ability to differentiate between the distinct groups as hypothesized is encouraging. It should, however, be noted that samples were very small so that effects of age and gender were not included in analyses. In addition, the groups were too small to be representative. The recruitment method among patient associations could reveal, in particular, children who were either more motivated, more limited or more knowledgeable about their illness. Results of hypotheses testing are therefore only indicative. Reassessment of differences between more representative groups is needed after revisions have been made in the questionnaire.

4.17.5 Concurrence between the child version and parent version

Moderate correlations between the child and the parent version proved that both versions should be used complementarily (Rosenbaum, Cadman Kirpalani, 1990; Achenbach, McConnaugh & Howell, 1987; LaGreca, 1990). The fact that the correlations were weaker for the social activity scales is understandable since an important part of a child's daily social activities is difficult for the parents to observe. The correlation between parents and children was higher on the prevalence scales than on the quality of performance scales, an explanation for this may be that the quality of performance scales are more dependent on individual judgement. In contrast to what was expected, the concordance on physical complaints was relatively high. This might be explained by the fact that in this age group it is especially parents have to help the child cope with physical complaints.

Parents were less optimistic than their children about the quality of performance of their children. Other authors also noted that proxies were less optimistic than the patients themselves (Tyc, Mulhern, Fairclough, Ward, Relling & Longmire, 1993; Sprangers & Aaronson, 1992).

4.17.6 Concluding remarks

As described above, revisions of items and answer categories in the generic section of the HAY are needed. In addition, an extensive revision is needed in respect of the chronic illness and disease-specific sections. It appeared that the chronic illness scales and disease-specific scales have to be adapted to capture fully the illness-specific issues of relevance. The scales measuring the prevalence of emotions towards illness, need more answer categories than a simple yes-no answer. In addition, a disease-specific daily management dimension is needed for children with asthma and children with epilepsy. Furthermore, the disease-specific physical complaints dimension needs further improvement for all four groups.

In respect of the methods used, larger sample sizes are needed to be able to obtain reliable results on validity and reliability of the generic as well as the disease-specific sections of the HAY. Multi-sample analyses have to be performed to assess the stability of the structure in all distinct groups. Revisions as described above are needed to obtain a version that is germane for children and parents.

ACKNOWLEDGEMENTS

The author would like to thank all the children and parents who participated in the study. Without the help of the Epilepsie Vereniging Nederland, de Reumabond en de Diabetes Vereniging Nederland, the two primary schools and the physicians who cooperated in the research, this study would not been possible.

CHAPTER 5: THE VALIDITY AND RELIABILITY OF THE HAY

5.1 INTRODUCTION

As was described in Chapter 4, the pilot version of the HAY required revisions following its trial. The reliability and validity of the revised instrument were reexamined in the main study. The main study consisted of a measurement among a large sample of children with asthma, insulin dependent diabetes mellitus, epilepsy, juvenile chronic arthritis and healthy children. In this chapter the results of the assessment of the validity and reliability of the revised version of the HAY are described.

5.2 RESEARCH QUESTIONS AND HYPOTHESES

The following research questions will be addressed in this chapter:

1. *What is the utility of the HAY?*
2. *What are the internal structure of the HAY and the reliability of dimensions?*
3. *Is the structure of the HAY stable in all distinct groups?*
4. *What is the concurrence of the HAY dimensions with other measures?*
5. *What is the discriminative power of the HAY?*
6. *What is the sensitivity of the HAY for severity of illness related symptoms?*
7. *What is the concurrence between child report and parent report?*

To assess utility, parents were asked whether their child filled in the questionnaire all by themselves. To assess the internal structure and reliability of the HAY, Confirmatory Factor Analysis (CFA) was performed to assess the fit of the predefined model and the Cronbach's alpha of the dimensions was calculated. In addition, multi-sample analyses were performed to assess the stability of the model in each group.

To assess the concurrence of the HAY with other measures, hypotheses were formulated relating to the correlation of dimensions of the HAY with dimensions of other measures. Besides the HAY, children answered the Child Attitude Toward Illness Scale (Austin & Huberty, 1993), which was hypothesized to correlate with the scales of the HAY measuring the prevalence of feelings of inferiority related to illness and the prevalence of concerns about the illness. The parents completed the validated Dutch version of the Child Behavior Check List (CBCL) (Verhulst, Koot, Akkerhuis & Veerman, 1990; Verhulst, Ende & Koot, 1996). Statistically significant

correlations were expected between scales of the CBCL measuring physical, social and cognitive competence, social problems and somatic complaints and the scales of the parent version of the HAY measuring respectively the quality of performance on physical activities, cognitive tasks and social activities, and the prevalence of social problems and physical complaints.

To assess the discriminative power of the HAY, hypotheses concerning the ability to differentiate between groups were formulated. It was hypothesized that children with a chronic illness would score lower on physical, social and psychological functioning (implying a lower prevalence and lower quality of performance levels of relevant behaviors and a higher prevalence of complaints) than healthy children according to child as well as parent report.

Based on the review of clinical studies among children with a chronic illness (see Chapter 1 and 2), it was concluded that age as well as gender are important variables to take into account when analyzing the results, as it has often been reported that boys experience more social and behavioral problems and that older children experience more problems in general. Girls are reported to have a lower physical competence and a lower self-esteem, seeing themselves as less attractive than boys do. In addition, girls are reported as experiencing more depression and more physical complaints. In order to assess differences between groups, differences were assessed separately for boys and girls while controlling for age.

Based on the review of studies described in Chapter 2, hypotheses concerning differences between each of the disease-specific groups and healthy children were formulated. Although most studies on which the hypotheses are based used parent reports to assess the functioning of children the following hypotheses are formulated to test differences between groups using parent report as well as child report.

It was hypothesized that *children with asthma* would experience more physical complaints and more limitations in physical activities as well as in social activities and would have more school absenteeism compared to healthy children. It was hypothesized that *children with diabetes mellitus* would have more problems in social functioning than healthy children. It was hypothesized that *children with epilepsy* would experience more limitations on cognitive tasks and social functioning than healthy children. Finally, it was hypothesized that *children with juvenile chronic arthritis* would experience more physical complaints and limitations in physical activities and social functioning and would have more school absenteeism than healthy children. As there were no clinical studies found on feelings in general or emotional problems related to illness, no hypotheses were formulated on these issues and assessment would be explorative. In addition, differences between each of the diagnostic groups compared to the other diagnostic groups were assessed exploratively.

Hypotheses concerning severity are not easy to formulate, as clinical studies indicate no linear relationship between severity and health-related quality of life (see Chapter 1 and 2). It might however be hypothesized that within a specific diagnostic group more symptoms or a higher severity of symptoms correlates with more limitations in physical and social activities, and cognitive tasks.

With regard to the concurrence between parents and children, it was hypothesized that there was a moderate correlation between parent and child report. It was expected that especially on objective issues that are easy to observe the correlation would be higher than the correlation on more subjective issues.

5.3 SAMPLE

The subjects in the main study at baseline were 1,038 children and 1,036 of their parents. For the recruitment of children with a chronic illness, six academic hospitals, 21 general hospitals, three specialized institutes, six patient organizations and several GPs cooperated. For the recruitment of healthy children, nine schools cooperated. In addition, a small number (56) of healthy children were recruited via their GP3. As a consequence of the relatively high prevalence of asthma, a considerable proportion of children with asthma was recruited through their schools (29 children).

Of the total group, 41 (4%) children and their parents were excluded from analyses leaving 995 children and parents in the study. Nine children who were initially included as healthy children, but whose parents indicated that they had a major chronic illness and used medication for this illness were subsequently excluded. In addition, thirteen children with a chronic illness were deleted from analyses because of comorbidity of their illness with other major chronic illnesses. Children of whom parents reported comorbidity of illnesses like eczema, food allergies or psychosocial problems were not excluded (see paragraph 5.8).

In addition, 19 children were excluded from analyses because of their age. Four children were considered to be too young, below seven years of age, to answer the questionnaire on their own. Fifteen children were considered to be too old, being 14 years or older, because issues included in the questionnaire were chosen in particular for primary school children from 7 up to 13 years old.

³ This part of the study was conducted by the EMGO Institute, VU Amsterdam (le Coq, 1998).

5.4 PROCEDURE

Parents of children with a chronic illness from all over the Netherlands were contacted by the child's GP, pediatrician or by a patient association. Children with a chronic illness were recruited through their parents. The reference group of healthy children and their parents were recruited through nine schools scattered throughout the Netherlands.

Parents and children were reassured that the data would be treated confidentially and that (non-) cooperation would not influence treatment of the chronic illness. Written informed consent was obtained from the parents. Two hospitals also asked for an informed consent from the children.

All children and parents were asked to complete the questionnaire separately from each other at home and to send it back to the researcher in a reply-paid envelope. Children were sent a small present as an incentive fee for their participation. Parents were promised a general report of the results of the study after the study was finished. Parents and children were asked to comment on items or their illness at the end of the questionnaire.

5.5 INSTRUMENTS USED IN THE STUDY

5.5.1 Description of the HAY

As described in Chapter 4, changes were made to the pilot version of the questionnaire. As in the pilot version, items refer to the past seven days except for the general treatment scale included in the parent's questionnaire. Figure 5.1 presents the revised version of the HAY.⁴

The generic section assesses the child's functioning in daily life areas, which are relevant for all children. This section includes eight dimensions: 1) physical activities, 2) cognitive tasks, 3) social activities, 4) social problems, 5) general physical complaints, 6) general treatment characteristics, 7) positive emotions, and 8) negative emotions. The physical activities, cognitive tasks and social activities (dimensions 1 to 3) are examined by asking the child about the prevalence of relevant daily activities, the quality of performance on the activities and the importance of the tasks/activities (assessed as feelings related to problems in the quality of performance of tasks/activities). Examples of items are respectively: (a) "Have you ridden your bike during the past seven days?" (b) "How well were you able to ride your bike during the past seven days?" and (c) "Does it bother you if you have a hard time riding your bike?" Social problems (dimension 4) and physical complaints (dimension 5) are measured by

⁴ As an example, the questionnaire for children with asthma is included in the Appendix. Versions with other disease-specific sections as well as the parent version, can be requested by the author.

asking about the prevalence of difficulties and the child's related feelings. Examples of items on social problems are: (a) "Have you been teased during the past seven days?" (b) "Does it bother you when you are teased?" Examples of items on physical problems are: (a) "Have you had headaches during the past seven days?" (b) "Does it bother you when you have a headache?" Since data on the prevalence of general treatment characteristics (dimension 6) cover a period of three months, they can be more reliably obtained from the parents. The children are only asked about their related feelings. An example is: (a) "Does it bother you if you have to stay in the hospital?" The emotions of a child (dimensions 7 and 8) are measured by the prevalence of emotions. An example is: "Have you felt happy during the past seven days?"

Figure 5.1: Description of the revised version of the HAY, its sections, dimensions, and scales

		Scales			
		Prevalence ¹	Quality of Performance ²	Feelings ³	
HOW ARE YOU? (HAY)	GENERIC SECTION	Physical activities	X	X	X
		Cognitive tasks	X	X	X
		Social activities	X	X	X
		Social problems	X		X
		Physical complaints	X		X
		Treatment			X
		Positive emotions	X		
		Negative emotions	X		
	CHRONIC ILLNESS SECTION	Concerns	X		
		Feelings of inferiority	X		
	DISEASE-SPECIFIC SECTION⁴	Physical Complaints	X		X
		Treatment tasks	X		X

1. All prevalence scales include the answer categories never/sometimes/often/very often.
2. All quality of performance scales include the answer categories well/not so well/not well/not well at all.
3. All feelings scales include four faces ranging from a happy to a very sad face undertitled with the categories not very much/a little/quite a bit/a great deal.
4. The two dimensions "disease-specific complaints" and "daily management" are developed for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis.

The chronic illness section consists of two dimensions: 1) concerns related to having an illness and 2) feelings of inferiority because of having an illness. An example of an item assessing concerns is: "Have you worried about the future due to your [illness] during the past seven days?" An item example related to feelings of inferiority is: "Have you felt during the past seven days that you were different from other children because of your [illness]?"

The *disease-specific section* related to the distinct illnesses can only be answered by children with a specific disease and consists of two dimensions: 1) physical complaints related to the specific illness and 2) daily management. Examples of items concerning asthma-related physical complaints are (a) "Have you felt short of breath during the past seven days?" and (b) "Does it bother you when you feel short of breath?" Examples of items concerning daily management for children with asthma are (a) "Have you taken your asthma medication during the past seven days?" (b) "Does it bother you when you have to take your asthma medication?"

The disease-specific sections referring to complaints and treatment tasks are developed for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. Disease-specific sections for children with cancer and children with chronic renal failure have been developed but have not been tested in the current validation study.

A *similar questionnaire for parents* was developed. The parents are asked similar questions on the prevalence and quality of performance of their child's daily functioning. They are however not asked about their child's feelings but about their own feelings related to the problems that occur for their child. Along with the items described in the HAY parents are asked in the same questionnaire to rate the medical history, disease-specific symptoms, functional limitations, medication use and visits to medical practitioners made by their child.

5.5.2 The Dutch version of the Child Behavior Checklist (CBCL)

The Dutch version of the Child Behavior Checklist (CBCL) was used, which was described in Chapter 4 (Verhulst, Koot, Akkerhuis & Veerman, 1990; Verhulst, Ende & Koot, 1996). Since 1996, data can be scored with a program that generates a profile of subscale scores as well as a total competence score, a total score for internalizing behavior problems and a total score for externalizing behavior problems that can be compared with Dutch norms within distinct gender and age groups.

5.5.3 Child Attitude Toward Illness Scale (CATIS)

A translated version of the Child Attitude Toward Illness Scale (CATIS) (Austin & Huberty, 1993) was used during the main study. The CATIS was designed to provide a systematic assessment of how favorably or unfavorably children feel about having a

chronic physical condition. A study among children with asthma and epilepsy, aged 8-12 years, provided support for construct validity of the scale, good internal consistency and test-retest reliability (Austin & Huberty, 1993).

The original measure included 13 items. Four of these items asked for the intensity of feelings, the remaining nine items were items asking children to rate how often they had feelings that would indicate positive or negative feelings associated with having a specific chronic illness. As the CATIS consisted of one scale and all items had to be included in a Confirmatory Factor Analysis, items asking for intensity were reformulated to items asking for the prevalence of feelings. One repeated item with a reversal in the direction of the responses was therefore deleted. The resulting translated version included ten items assessing negative attitudes towards having the chronic illness and two items assessing positive attitudes. In contrast to the original measure, the five answer categories of items were set from “never” to “always”.

As the questionnaire was translated and adapted to the Dutch situation, a Confirmatory Factor Analysis was performed to assess the structure of the CATIS. Two items were deleted because of nonnormal distributions. A third item was deleted because of a low squared multiple correlation and a fourth item because of large standardized residuals. The fit of the resulting one-factor model of 8 items was good, the χ^2 was 107.05 (N=563, Df=20; p=.000), the GFI was .95, the AGFI=.91 and the RMR was .05, the total coefficient of determination was .89. The Cronbach’s alpha of the CATIS for all groups was .88 (see Table 5.1 for lambda values, item-total correlations and Cronbach’s alpha).

Table 5.1: Lambda-values, item-total correlations and Cronbach’s alpha of the translated version of the CATIS

Item	Lambda ^a	Item-total corr. ^b	Alpha ^c
How often do you feel terrible about having [x] ^d	.80	.67	.88
How often do you feel that it is unfair that you have [x]	.90	.66	
How often are you sad about having [x]	.91	.70	
How often is it difficult to do the things you want to do because of your [x]	.73	.58	
How often do you think you will always be ill	.67	.54	
How often do you feel different from other people because you have [x]	.66	.61	
How often do you feel fed up with having [x]	.86	.69	
How often do you feel sad about having an illness	.87	.72	

^a All lambda-values are significant at p < .002

^b Item-total correlations

^c Cronbach’s alpha

[x] The illness of the child is filled in, asthma, diabetes mellitus, epilepsy or juvenile chronic arthritis

5.6 DATA INPUT AND DATA ANALYSIS

The *data* derived from the HAY and the CATIS were entered by means of the Data Entry program of SPSS-PC+ (Norusis, 1988). The data derived from the CBCL were entered by means of the official program diskette and scored with the use of the Dutch norms (Verhulst, Ende & Koot, 1996). In both programs ranges were provided for checking valid values of each variable during input. Furthermore, a small selection of questionnaires was entered a second time (14 addresses), which revealed only a few minor typing errors. In addition, the data were reviewed by using descriptive statistics as Frequencies, Descriptives and Crosstabs.

Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations, a high level of positive emotions, a low level of negative emotions, a high quality of performance and a low level of negative feelings. Scores of items within each scale were summarized and averaged to be able to compare scores at different scales. Results of the confirmatory factor analysis on feelings were used to form related prevalence or quality of performance scales. The scales in the parent version were based on the results of analyses of the child version.

Differences in baseline characteristics of groups were tested by non-paired Student t tests and one-way analyses of variance in the case of more than two groups for continuous data and by Pearson chi-square tests of association for nominal data. For ordinal data, Kruskal-Wallis one-way analysis of variance by ranks was used. For significance of differences, a significance level of .05 was used.

Confirmatory factor analysis, goodness of fit and reliability. Before applying the confirmatory factor analysis, items were checked for relevance by analyzing the prevalence item distributions. Criteria for inclusion were that at least 60% of the children of the total group participated in physical and social activities and cognitive tasks during the past seven days and that difficulties such as having a physical symptom or a negative emotion occurred in at least 10% of the children. Secondly, items included in the confirmatory factor analysis were checked on normal distributions by assessing skewedness, kurtosis, and inspecting graphical plots.

As the HAY consists of separate sections, and is available for children with a chronic illness as well as for each of the disease-specific groups, the LISREL analyses had to be performed separately on the generic section, the chronic illness section, and the disease-specific section for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. As the analyses on the generic section had to be performed on related feelings items as well as on the prevalence of emotions, the structure of the prevalence of emotions had to be analyzed separately from the related feelings items.

In addition, *multi-sample analyses* were performed to test the stability of the factor structure among the distinct groups of the generic section of the HAY as well as the disease-specific emotional sub-section. For these analyses, LISREL 8 was used as LISREL 7 did not have enough working memory to perform the analyses (Jöreskog & Sörbom, 1989; 1993).

When items were normally distributed, covariance matrices were used (Jöreskog & Sörbom, 1989; Byrne, 1989). All covariance matrices used were assessed by PRELIS 1.13 and analyzed in LISREL 8.14 using the maximum likelihood method (ML). When, however, items were non-normal distributed, polychoric correlation matrices were assessed using PRELIS 1.13 and in LISREL the weighted least squares method (WLS) was used (Jöreskog & Sörbom, 1989). The WLS method generally requires a large sample size and is as a result considered to be asymptotically free (Schumacker & Lomax, 1996).

Assessment of the overall fit was based on the χ^2/df ratio, the goodness-of-fit index (GFI) and adjusted goodness-of-fit index (AGFI), the root-mean-square residual (RMR) and the total coefficient of determination provided by LISREL 7 (Jöreskog & Sörbom, 1989). To identify sources of misfit within a specified model, a more detailed evaluation of fit was obtained by inspecting the squared multiple correlation (R^2), the standardized residuals, t-values of the parameters and modification indices (MI) as well as the PHI values provided by LISREL as was described in Chapter 4.

Furthermore, *Cronbach's alpha* was used to assess the internal consistency of scales in the child version as well as parent version. When the reliability estimates of distinct scales revealed too low alphas in the child version (below .70), models were reassessed.

In the *multi-sample analyses* using LISREL 8.14 (Jöreskog & Sörbom, 1989), the comparative fitness index (CFI) and the root-mean-square-error of approximation (RMSEA) were used as these values are less dependent on sample sizes. The CFI represents an estimate of the relative amount of variance that is explained by the model when compared to the explanatory power of the independence model, estimating the relative difference in noncentrality of interest (Bentler, 1990). A CFI of .90 was considered to be moderately good, a CFI of .95 or higher as good. The RMSEA is a measure that takes into account the error of approximation in the population and the precision of the fit measure. A value of about 0.05 or less refers to a close fit of the model in relation to the degrees of freedom, a value between 0.05 and 0.08 indicates a reasonable error of approximation and a model with a RMSEA of 0.10 or more was not considered to be acceptable (Browne & Cudeck, 1992).

Especially in the multi-sample analyses, small sample sizes have occurred within the groups studied. With small samples, the values of the test statistics are frequently too

large leading to a rejection of correct models. However, one can rely on the test procedure unless the samples become very small and/or the variables have very skewed distributions (Saris & Stronkhorst, 1984).

Concurrence with other instruments was assessed using Spearman Rank correlations.

In order to establish the *discriminative power of the HAY*, differences between groups were established using General Linear Models (GLM) (SPSS) to perform a two-factorial analysis of variance (ANOVA) with gender and illness as independent factors and age as covariate. This was done for the comparisons between healthy children and children with a chronic illness as well as for studying the differences between the distinct groups. Furthermore, the differences between the five groups were examined in detail by using contrasts, method=simple, in which each level, except the last, of the factor illness is compared to the last level. Because of multiple testing, an alpha level of .01 was used. When the scales were non normally distributed, a transformation of the scale was used for analyses of variance ($X_{\text{new}}=1/(5-X_{\text{old}})$) (Tabachnik & Fidell, 1989, Stevens, 1992). In the tables, non-transformed variables are printed to be able to interpret the findings. The Levene's test for homogeneity of error variances was used to test the *significance of differences in variance* (Stevens, 1992). When the Levene's test was significant ($p<.01$) and the largest sample variances were associated with the smallest group sizes, the F statistic was considered to be liberal. To compensate this, a significance level of .001 was used in these cases (Stevens, 1992).

In order to assess the *sensitivity* of dimensions of the HAY for current disease activity within each group, parent-reported current disease activity was related to prevalence and quality of performance scales of the HAY. Hierarchical multiple regressions were conducted to test the relative contribution of current disease-activity after controlling for children's age and gender. The alpha was set on .01 for all tests of regression coefficients.

Concurrence between parents and children. The concurrence between scores of children and parents at individual level was established by intra-class correlations (ICC) and Pearson's correlation or Spearman Rank correlations in case of non-normal distributions of scores. Paired Student t tests were used to compare mean scores of parents and children to assess whether there was a systematic bias in reporting quality of life. When the scales were non-normally distributed, a transformation of the scale was used for analyses of variance ($X_{\text{new}}=1/(5-X_{\text{old}})$) (Tabachnik & Fidell, 1989, Stevens, 1992). The ICC is the proportion of total variability accounted for by the variability among persons only. The ICC combines information from the t test and product-moment correlation and ranges from 0 to 1. If it is high, it means that not much of the variability is due to variability in measurements by either parents or children, or put in another way, the answers are interchangeable. The ICC is calculated on the basis of results of paired t-tests in the group of healthy children

and children with a chronic illness. The formula given by Deyo, Diehr and Patrick (1991) was used.

5.7 BASELINE CHARACTERISTICS OF THE SAMPLE

In total 995 children and their parents were included in the study, of whom 577 children had a chronic illness and 418 were healthy children.

Table 5.2: Demographic characteristics of the research population

Variable	Category	Ill ¹ (n=577)	Healthy ¹ (n=418)
Mean age children (SD) and range		10.35 (1.7) 7.0-13.9	10.27 (1.2) 7.2-13
Gender	N boys	291(50%)	201 (48%)
	N girls	286(50%)	217 (52%)
Family composition:			
One or two parent families	Two parent	520 (90%)	380 (91%)
	One parent	50 (9%)	31 (7%)
Number of children within families	1 child	75 (13%)	33 (8%)
	2 children	265 (46%)	219 (53%)
	3 children	157 (27%)	122 (29%)
	>3 children	73 (13%)	41 (10%)
Respondent parent	Mother	504 (88%)	346 (84%)
	Father	50 (9%)	62 (15%)
	Other ²	19 (3%)	6 (1%)
Mean age respondent parent (SD)		38.69 (4.3)	39.45 (4.2)
Educational level respondent parent ³	Lower	191 (33%)	145 (35%)
	Intermediate	182 (32%)	125 (30%)
	Higher	142 (25%)	96 (23%)
	Other	54 (9%)	50 (12%)
Nationality respondent parent	Dutch	550 (95%)	411 (98.8%)
	Other European	11 (1.9%)	1 (0.2%)
	Other	13 (2%)	4 (1%)

¹ Due to missing values, total numbers do not add up to 995

² In most cases this means both parents together

³ Lower educational level=elementary education, MAVO (general secondary education-junior level), VBO (lower vocational education); intermediate educational level=HAVO/VWO (general secondary education-senior level) and MBO (vocational education-junior level); higher educational level=HBO (vocational education-senior level) and WO (university education)

Children who participated in the study were healthy children and children who had been diagnosed with asthma, diabetes mellitus, epilepsy or juvenile chronic arthritis. Other criteria for selection were that the children with a chronic illness had made a

visit to the pediatrician or general practitioner at least once in the last year, that they had no more than one chronic illness and that they were not hospitalized when the questionnaire was administered. In addition, children had to attend normal primary schools. Furthermore, an important requirement was that all children and parents were able to read Dutch.

There were no statistically significant differences ($p < .05$) in gender or age between the two groups of children ($p < .05$). Nor were there statistically significant differences in the percentages of one or two parent families, number of children, nationality or educational level of the respondent parent. However, statistically significant differences existed for age and gender of the respondent parent ($p < .05$). The group of parents of healthy children included more fathers and had a higher mean age than the group of parents of children with a chronic illness.

The group of children with a chronic illness is divided into 275 children with asthma, 139 children with diabetes mellitus, 104 children with epilepsy and 59 children with juvenile chronic arthritis. There were no significant differences found for age between the groups. As expected from the prevalence estimates described in Chapter 1 and the etiology described in Chapter 2, statistically significant differences were, however, found for gender ($p < .05$), illness duration ($p < .05$) and illness onset ($p < .05$) between the four distinct groups.

Table 5.3: Demographic characteristics of the distinct illness groups

	Category	Asthma	IDDM	Epilepsy	JCA
Total number		275	139	104	59
Gender \bar{n} (%)	Girls	114 (41%)	71 (51%)	57 (55%)	44 (75%)
	Boys	161 (59%)	68 (49%)	47 (45%)	15 (25%)
Mean age (SD)		10.2 (1.6)	10.7 (1.8)	10.3 (1.8)	10.4 (1.7)
Range		(7.2-13.9)	(7.3-13.9)	(7.0-13.9)	(7.3-13.9)
Mean age at onset (SD)		3.8 (2.83)	6.9 (2.6)	7.5 (2.6)	5.7 (1.7)
Range		(.02-12.5)	(1.2-12.3)	(0.50-11.7)	(.8-12.52)
Mean illness duration (SD)		6.3 (3.0)	3.8 (2.7)	2.9 (2.8)	4.6 (2.6)
Range		(0.2-12.8)	(0-12.0)	(.08-12.7)	(.3-10.7)

The group of children with asthma included more boys and the group of children with juvenile chronic arthritis included more girls than the other groups ($p < .05$). Parents of children with asthma reported a significant longer duration of illness, and a younger age of onset than parents of the other groups did ($p < .05$). Parents of children with juvenile chronic arthritis reported a younger age at onset and a longer disease duration than parents of children with epilepsy reported ($p < .05$).

5.8 COMORBIDITY, LIMITATIONS AND GENERAL TREATMENT

In this paragraph a more detailed description of the distinct illness groups will be given including a description of comorbidity of illnesses, limitations and use of medical care as reported by the parents.

5.8.1 Comorbidity

Parents were asked in an open-ended item whether their child suffered from additional chronic illnesses. In Table 5.4, the comorbidity of illnesses is presented.

Statistically significant differences were found in the number of children with comorbidity as reported by the parents ($p < .05$). A relatively large proportion of children with asthma (28%) have other illnesses, mainly eczema, and allergies. Ten percent of the children with epilepsy were reported to have comorbidities, such as eczema, allergies and hyperactivity, whereas for the groups of children with diabetes mellitus, juvenile chronic arthritis and the group of healthy children respectively 3%, 3% and 1% comorbidities were reported.

Table 5.4: Comorbidity of illnesses

	Asthma ¹ n (%)	IDDM n (%)	Epilepsy n (%)	JCA n (%)	Healthy n (%)
Food allergy	8 (4%)				1
Eczema	26 (13%)		1 (1%)	2 (3%)	
Allergy	13 (7%)	3 (2%)	2 (2%)		4 (1%)
Bronchitis	3 (2%)		2 (1%)		1
Skin disease	1 (1%)	1 (1%)	1 (2%)		
Psychological problems	1 (1%)		1 (1%)		
Ear problems	3 (2%)		1 (1%)		
Hyperactivity			2 (2%)		
Total of children with comorbidity	55 (28%)	4 (3%)	10 (10%)	2 (3%)	6 (1%)
Total number	193	139	97	59	415

¹ Results are missing from part of the sample that was provided by the EMGO Institute

5.8.2 Limitations

The parents were asked to rate on a four point scale the limitations their children experience in following their daily activities. As expected from the prevalence estimates described in Chapter 1, there is a significant difference in the degree of limitations children of the distinct groups experienced according to their parents ($p < .001$). Children with asthma and children with juvenile chronic arthritis experienced most limitations in doing their daily activities. As was expected, healthy children were reported to experience fewest limitations.

Table 5.5: *Limitations encountered by children according to their parents*

Category	Healthy n (%)	Asthma n (%)	IDDM n (%)	Epilepsy n (%)	JCA n (%)
Severely hindered	4 (1%)	71 (26%)	1 (1%)	0	2 (4%)
Considerably hindered	4 (1%)	11 (4%)	4 (3%)	2 (2%)	9 (16%)
Somewhat hindered	51 (12%)	84 (31%)	25 (18%)	20 (19%)	22 (39%)
Not at all hindered	354 (86%)	104 (39%)	109 (78%)	81 (79%)	24 (42%)
Total number of children ¹	n=413	n=270	n=139	n=103	n=57

¹ Because of rounding off, columns do not add up to 100%

5.8.3 Medical care

All children with a chronic illness are, to a greater or lesser extent, dependent on medical care. In Table 5.6 in the appendix, the number of visits due to the chronic illness which are paid during the past six months to professional health carers including specialists, GPs and others are described.

A relatively low percentage of children with asthma had consultations with the specialist (77% compared to 90% or more for the other groups). Although many children with asthma can be treated by the GP, this larger percentage may also be caused by our recruitment method as part of the children with asthma was recruited in GP practices. A relatively large percentage of children with asthma had attended the GP during the past six months for treatment of their asthma. A relatively large proportion of children with diabetes mellitus and children with juvenile chronic arthritis had spent one or more days in hospital. A relatively large proportion of children with juvenile chronic arthritis is dependent on other health carers, such as a physiotherapist (see Table 5.6 in the Appendix).

5.9 UTILITY OF THE HAY

As the majority of the questionnaires were sent to parents and children at home, the parents were asked to indicate whether the child had filled in their questionnaire independently. According to the parents, most children (N=855) (89%) answered the HAY alone. Of the group of 7 to 8 year olds, 49 children (5%) did not fill in the HAY all by themselves according to the parents; of the group 9-10 year olds, 30 children (3%) did not fill in the HAY alone; of the group 11-12 year olds 18 children (2%) did not fill in their questionnaire alone and only four of the 13-year olds did not fill in their questionnaire alone.

5.10 INTERNAL STRUCTURE AND RELIABILITY OF THE GENERIC SECTION OF THE HAY

5.10.1 Inspection of item distribution

Before conducting confirmatory factor analysis, the prevalence items were checked for relevance by analyzing the prevalence item distributions. All the prevalence items measuring general emotions were within the 10% relevance interval except for two items concerning going to parties and having dinner with friends. Nevertheless, these items were included because the children with a chronic illness saw them as relevant according to the results of the pilot study.

Secondly, the related feelings items and emotional items on which the confirmatory were conducted were assessed on normality of distributions by examining the skewedness, kurtosis and graphical plots. The related feelings scales and general positive emotion items of the generic part of the HAY were normally distributed. The negative emotion items of the generic part of the HAY were however highly skewed and leptokurtic. Confirmatory factor analysis had thus to be conducted using polychoric correlations and the weighted least squares method (Saris & Stronkhorst, 1984; Jöreskog & Sörbom, 1989).

5.10.2 Results of confirmatory factor analysis and reliability testing

LISREL analysis with the six generic scales revealed a moderately good fit for the six factor solution. The value of χ^2 (N=991, Df=390, p=.000) was 1314.63, the goodness of fit (GFI) was .91, adjusted goodness of fit (AGFI) was .90 and the root-mean square residual (RMR) was .05. The total coefficient of determination was 1.0. Low squared multiple correlations (lower than .2) were, however, found for three items and relatively high multiple modification indices (MI) were found for eight items. These eleven items were deleted. Cronbach's alphas of all scales, except the general treatment scale ($\alpha=.65$), was higher than .70. The treatment scale was therefore deleted.

Further analyses revealed that a better fit was obtained for a four-factor solution including 17 items assessing physical activities, cognitive tasks, social activities and physical complaints. The value of χ^2 was 411.28 (N=991, Df=113, p=.000), GFI was .95, AGFI was .93 and RMR was .04. The total coefficient of determination was 1.0. The lambda values, which can be seen as factor loadings, ranged from .46 to .77 and were significant at a level of $p < .001$. As most fit indices were good, this model was accepted. The Cronbach's alphas of the first four scales in the child version ranged from .77 to .83. The Cronbach's alphas of the separate scales in the parent version ranged from .86 to .93 (see Table 5.7 for lambda values, item-total correlations and Cronbach's alphas).

Table 5.7: Lambda values, corrected item-total correlations and Cronbach's alphas for the generic section of the *How ARE YOU? (HAY)*

Scale Item ¹	Lambda ^a	Item-total corr. ^b	Alpha ^c	Item-total corr. ^d	Alpha ^e
I. Physical activities			.83		.88
Does it bother you when you					
have trouble biking	.65	.66		.70	
have trouble playing outside	.71	.65		.70	
have trouble running	.64	.61		.78	
have trouble joining in gym	.66	.61		.74	
have trouble swimming	.66	.59		.70	
II. Cognitive tasks			.81		.92
Does it bother you when you	.70	.64		.85	
have trouble doing math assignments	.64	.67		.82	
have trouble concentrating	.73	.71		.88	
have trouble doing language assignments	.57	.52		.70	
can't remember what you learned at school					
III. Social activities			.77		.86
Does it bother you when	.45	.48		.67	
you are not able to go to a party	.70	.58		.72	
playing with other children during recess	.72	.60		.72	
playing with other children after school	.43	.47		.61	
you are not able to have diner at a friend's	.53	.55		.64	
you are not able to pay a visit to someone					
IV. Physical complaints			.83		.93
Does it bother you when you	.55	.64		.79	
have a headache	.59	.70		.88	
are nauseous	.62	.72		.88	
have a stomach ache					
V. Happiness			.86		.89
Did you feel happy	.55	.66		.72	
Have you felt cheerful	.61	.72		.79	
Did you laugh	.57	.65		.79	
Did you have fun	.62	.72		.60	
Have you felt pleased	.56	.65		.76	

¹ The parents' own feelings are assessed in the parent version

^a All lambda values are significant at $p < .002$

^b Item-total correlations in the child version

^c Cronbach's alphas in the child version

^d Item-total correlations in the parent version

^e Cronbach's alphas in the parent version

^f Full questionnaire is added in the Appendix

CFA using polychoric correlations revealed that the two-factor model of the emotional dimensions in the generic section had a good fit, χ^2 was 59.94 (N=725, Df=26, $p=.000$)⁵, GFI was .99, AGFI was .99, RMR was .05. The total coefficient of determination was .98. The PHI value was .31. Assessment of the internal consistency of the positive and negative emotional dimensions revealed however a low Cronbach's alpha of the negative emotions scale, which was .67. Therefore the two-factor model was rejected.

A one-factor model including only the positive emotions was reassessed on the basis of a covariance matrix as these items were normally distributed. The one-factor model had a good fit, χ^2 was 45.34 (N=731, Df=5, $p=.000$), GFI was .98, AGFI was .93 and RMR was .02. The total coefficient of determination was .86. Cronbach's alpha in the child version was .86. One item had to be deleted because of a low contribution to Cronbach's alpha. The final scale was called "happiness". Cronbach's alpha in the parent version was .89. In Table 5.7 the generic section of the HAY is presented.

Table 5.8 represents the correlations between the latent factors (PHI values) and Pearson correlations between the first four dimensions among respectively the group of healthy children (a) and the group of children with a chronic illness (b).

Table 5.8: PHI values and Pearson correlations among (a) healthy children, (b) children with a chronic illness

(a) Healthy children		PHI values				Pearson r			
Factor I-IV		I	II	III	IV	I	II	III	IV
I	Physical functioning	1.00				1.00			
II	Cognitive functioning	.75	1.00			.63*	1.00		
III	Social functioning	.78	.64	1.00		.59*	.48*	1.00	
IV	Physical complaints	.31	.34	.40	1.00	.27*	.29*	.36*	1.00

(b) Children with a chronic illness		PHI values				Pearson r			
Factor I-IV		I	II	III	IV	I	II	III	IV
I	Physical functioning	1.00				1.00			
II	Cognitive functioning	.65	1.00			.55*	1.00		
III	Social functioning	.69	.51	1.00		.53*	.42*	1.00	
IV	Physical complaints	.39	.32	.44	1.00	.33*	.28*	.38*	1.00

* $p < .01$

The correlations between the latent factors (PHI values) of the first three latent factors were fairly high in the healthy group, ranging from .31 to .78, but acceptable in the chronic illness group, ranging from .32 to .69. To decide whether the latent

⁵ As the dimension measuring positive emotions was somewhat later included in the HAY than the other dimensions and the version used by the EMGO Institute didn't include this dimension, data from 148 children with asthma and their parents and 115 healthy children and their parents on this dimension are missing.

factors were independent, the Pearson correlations should be substantially lower than the mean Cronbach's alpha. Examination of the Pearson correlations and mean Cronbach's alpha of the dimensions revealed that these three factors represented enough unique variance. The dimension happiness was not included in these analyses as this dimension was examined separately from the first four dimensions as was described above.

Multisample analysis revealed that the four-factor structure was stable in the five distinct groups. When testing the hypothesis that factor loadings, factor correlations and error variances were identical in all groups, χ^2 was 1222.67 (N=991, Df=725, $p=.000$), CFI was .92 and RMSEA was .03. These goodness-of-fit indices together indicated a good fit.

Multisample analysis to test the hypothesis that the factor loadings and error variances of the happiness factor were invariant between the five groups, revealed that the fit was good: χ^2 was 118.28 (N=731, Df=65, $p=.000$), CFI was .97 and RMSEA was .03. These fit indices together indicated a good fit.

As explained, the LISREL analyses on the first four dimensions were conducted on the feelings items. The resulting dimensions were used to form the corresponding prevalence scales and quality of performance scales, so that the physical (5 items) and social activities (5 items) dimensions and the cognitive task dimension (4 items) each consists of three scales, measuring the prevalence, quality of performance and feelings towards limitations in performance. The physical complaints dimension consists of two scales each of three items, the prevalence of physical complaints and feelings towards physical complaints. The happiness dimension consists of just one scale, the prevalence of happy emotions, including 5 items. This resulted in a generic section of a total of 53 items.

5.11 INTERNAL STRUCTURE AND RELIABILITY OF THE CHRONIC ILLNESS SECTION

This section consists of items referring to emotions related to illness and is applicable to all children with a chronic illness. As described above, it was hypothesized that this section consisted of two latent factors: the prevalence of concerns related to having a specific illness and the prevalence of feelings of inferiority because of having a specific illness.

Item distributions indicated that these items were non-normally distributed, having a high skewedness and kurtosis, so that matrices containing polychoric correlations and the WLS method in LISREL were used.

The assessment of the two-factor solution revealed a very good fit. The value of χ^2 was 21.49 ($N=563$, $Df=26$, $p=.716$), GFI was 1.0, AGFI was .99, and RMR was 0.04. The total coefficient of determination was .97. The lambda values ranged from .72 to .86 and were significant at a level of $p < 0.001$ (see Table 5.9).

The correlation between the latent factors (PHI values) was .82, the Pearson correlation between the scales was .61. Since the PHI value was high, a one-factor model with all items included, except for one item because of a low squared multiple correlation, was tested and was rejected because of a poorer fit. The value of χ^2 of this one-factor model was 60.56 ($N=563$, $Df=27$, $p=.000$), GFI was .99, AGFI was .98 and RMR was 0.06. The total coefficient of determination was .93.

Table 5.9: Lambda values, corrected item-total correlations and Cronbach's alphas for the chronic illness section of the HOW ARE YOU? (HAY)

Scale Item	Lambda ^a	Item-total corr. ^b	Alpha ^c	Item-total corr. ^d	Alpha ^e
I Concerns about the illness¹			.80		.86
Have you worried about your [x]	.80	.60		.75	
Were you scared about becoming [x-symptom]	.75	.57		.67	
Have you felt sad because of your [x]	.84	.60		.73	
Have you felt angry because of [x]	.79	.57		.57	
Have you worried about your future because of your [x]	.75	.57		.64	
II Feelings of inferiority because of having a specific illness			.74		.66
Have you felt lonely because of your [x]	.86	.56		.40	
Have you felt that other kids did not like you as much because of [x]	.72	.47		.37	
Have you felt that you were not able to do as much as other kids [x]	.74	.52		.45	
Have you felt that you were different from other kids because of your [x]	.85	.57		.52	

Note. Full chronic illness section is printed in the Appendix. [x] = illness. The parents' own feelings are assessed in the parent version

^a All lambda values are significant at $p < .002$

^b Item-total correlations, child version

^c Cronbach's alphas, child version

^d Item-total correlations, parent version

^e Cronbach's alphas, parent version

The two-factor model had thus a better fit. The Cronbach's alphas were .80 for the scale measuring "concerns about the illness" and .74 for "feelings of inferiority because of having a specific illness" (Table 5.9) in the child version. The mean of the Cronbach's alphas was substantially higher than the Pearson r , indicating that the two factors represented enough unique variance. The two-factor model was thus accepted. The Cronbach's alphas of the parent version were .86 for the scale

measuring “concerns about their child’s illness” and .66 for “parents’ feelings of inferiority because of their child having a specific illness” (Table 5.9).

As polychoric correlation matrices need large sample sizes, multi-sample analyses were not possible using polychoric correlation matrices. The results of the LISREL analysis using a covariance matrix however revealed minor differences with the results of the analysis performed using polychoric correlations, so that multi-sample analyses were therefore performed on covariance matrices.

Multi-sample analysis revealed that the two-factor model was stable in the four distinct groups. When testing the hypothesis that factor loadings, factor correlations and error variances were identical in all groups, χ^2 was 601.52 (N=563, Df=161, $p=.000$), CFI was .74 and RMSEA was .07. Testing the hypothesis that the structure was identical in all groups but the factor loadings, factor correlation and error variances were free, χ^2 was 258.73 (N=563, Df=104, $p=.000$), CFI was .91 and RMSEA was .05 indicating a reasonable error of approximation. The disease-specific sub-section of the HAY referring to emotions related to illness includes 9 items in total.

5.12 DISEASE-SPECIFIC SECTION FOR CHILDREN WITH ASTHMA

Table 5.10: *Lambda values, corrected item-total correlations and Cronbach’s alphas for the asthma-specific section of the HOW ARE YOU? (HAY)*

Scale Item	Lambda	Item-total corr. ^b	Alpha ^c	Item-total corr. ^d	Alpha ^e
I. Physical complaints			.77		.83
Does it bother you when you					
- get up feeling tired	.46	.47		.52	
- have to cough	.52	.52		.66	
- are short of breath	.59	.62		.69	
- are wheezy	.61	.59		.66	
- wake up at night due to your asthma	.50	.50		.64	
II. Daily management			.76		.78
Does it bother you when you					
- have to take medication for your asthma	.66	.57		.56	
- have to take your asthma medicine with you	.72	.63		.70	
- have to use your peak flow measure	.47	.50		.53	
- have to take your asthma medicine when other children are around	.53	.52		.56	

Note. For full asthma-specific section see the Appendix. The parents’ own feelings are assessed in the parent version.

^a All lambda values are significant at $p < .002$

^b Item-total correlations, child version

^c Cronbach’s alphas, child version

^d Item-total correlations, parent version

^e Cronbach’s alphas, parent version

As described above it was hypothesized that the disease-specific section for children with asthma consisted of the dimensions, “asthma-related physical complaints” and “daily management”. This model indicated a good fit. After the deletion of one item because of too low a squared multiple correlation (smaller than .2), χ^2 was 43.60 (N=240, Df=26, p=.02), GFI was .96, AGFI was .94 and RMR was .04. The total coefficient of determination was .95. The PHI value between the factors was .40. Cronbach’s alphas of these scales in the child version were respectively .77 and .76. Cronbach’s alphas of these scales in the parent version were respectively .83 and .78. In Table 5.10 the results of confirmatory factor analysis and reliability testing are presented.

As explained, the LISREL analyses were conducted on the feelings items. The resulting dimensions were used to form the corresponding prevalence scales, so that the asthma-related physical complaints dimension consists of two scales each of 5 items, the first one measuring the prevalence of physical complaints and the second one measuring related feelings towards physical complaints. The daily management dimension consists of two scales each of 4 items measuring respectively the prevalence of daily management and the feelings towards daily management. The asthma-specific section consists of 18 items in total.

5.13 DISEASE-SPECIFIC SECTION FOR CHILDREN WITH DIABETES MELLITUS

As described above, it was hypothesized that the disease-specific section for children with diabetes mellitus consisted of the factors, “diabetes mellitus related physical complaints” and “daily management”. This model indicated however an unsatisfactory fit. The two-factor model had a χ^2 of 63.48 (N=134, Df=34, p=.002), GFI was .91, AGFI was .85, and RMR was .06. The total coefficient of determination was .90. Two items were deleted because of too low a squared multiple correlation (smaller than .2). Cronbach’s alphas of the scales were respectively .65 and .63.

Content analyses of the factors revealed that a three-factor model could reveal a better fit, namely “diabetes mellitus-related complaints related to a hypoglycemia” and “diabetes mellitus-related complaints related to a hyperglycemia” and “daily management”. The three-factor model indicated a good fit, χ^2 was 33.33 (N=132, Df=32, p=.403), GFI was .95, AGFI was .92 and RMR was .04. The total coefficient of determination was .94. The PHI value between the first and second factor was .55, between the first and third factor it was .56 and between the second and third .20. Cronbach’s alphas of the scales in the child version were however low, ranging from .58 to .63. Cronbach’s alphas of the scales in the parent version ranged from .62 to .71. In Table 5.11 the results of confirmatory factor analysis and reliability testing are presented

The LISREL analyses were conducted on the feelings items. The resulting dimensions were used to form the corresponding prevalence scales, so that the diabetes mellitus-specific sub-section consists of three dimensions of two scales each. The first dimension includes two scales measuring the prevalence of hyperrelated complaints and feelings towards these complaints (each of 4 items). The second dimension includes two scales measuring the prevalence of hypo-related complaints and the feelings towards these complaints (each of 3 items). The third dimension includes two scales measuring the prevalence of daily management and feelings towards daily management (each of 3 items). The diabetes mellitus-specific section consists thus of 20 items.

Table 5.11: *Lambda-values, corrected item-total correlations and Cronbach's alphas for the diabetes mellitus-section of the HOW ARE YOU? (HAY)*

Scale Item	Lambda ^a	Item-total corr. ^b	Alpha ^c	Item-total corr. ^d	Alpha ^e
I. Hyper related complaints					
Does it bother you when you			.58		.62
- have hyperglycemia in the daytime	.50	.44		.38	
- don't feel fit	.48	.38		.44	
- are not allowed to eat what you want	.39	.31		.24	
- have bruises from insulin injections	.34	.30		.51	
II. Hypo related complaints					
Does it bother you when you			.63		.67
- are dizzy	.51	.43		.33	
- have hypoglycemia in daytime	.56	.47		.61	
- have hypoglycemia at night	.49	.43		.58	
III. Daily management					
Does it bother you			.63		.71
- when you have to take blood sugar tester with you	.30	.37		.43	
- when your blood sugar levels have to be tested	.49	.52		.68	
- that you need insulin injections	.62	.42		.50	

Note. The parents' own feelings are assessed in the parent version

For the full diabetes specific section, please contact the author

^a All lambda-values are significant at $p < .002$

^b Item-total correlations, child version

^c Cronbach's alphas, child version

^d Item-total correlations, parent version

^e Cronbach's alphas, parent version

5.14 DISEASE-SPECIFIC SECTION FOR CHILDREN WITH EPILEPSY

As described above, it was hypothesized that the disease-specific section for children with epilepsy consisted of the factors, "epilepsy-related physical complaints" and "daily management". This model indicated however an

unsatisfactory fit. The two-factor model had a χ^2 of 77.14 ($N=89$, $Df=34$, $p=.000$), GFI was .84, AGFI was .74, and RMR was .07. The total coefficient of determination was .92. Cronbach's alphas of the scales was respectively .80 and .63.

Table 5.12: Lambda values, corrected item-total correlations and Cronbach's alphas for the epilepsy-specific section of the HOW ARE YOU? (HAY)

Scale Item	Lambda ^a	Item-total corr. ^b	Alpha ^c	Item-total corr. ^d	Alpha ^e
I. Physical complaints					
Does it bother you when you			.74		.72
- get up feeling tired	.73	.67		.59	
- are dizzy	.52	.50		.56	
- feel sleepy	.57	.55		.45	
II. Seizures					
Does it bother you when you			.78		.83
- have a "grand mal" seizure	.61	.57		.54	
- have an episode of "not being quite there"	.79	.74		.84	
- experience any symptoms associated with epilepsy such as	.60	.53		.75	
III. Daily management					
Does it bother you when you			.70		.76
- have to take medication for your epilepsy	.51	.44		.38	
- have any trouble with your medicine for epilepsy	.73	.55		.55	
- have to take your epilepsy in mind when	.51	.44		.62	
- are not allowed to do something because you have epilepsy	.61	.53		.61	

Note. The parents' own feelings are assessed in the parent version

^a All lambda values are significant at $p < .002$

^b Item-total correlations, child version

^c Cronbach's alphas, child version

^d Item-total correlations, parent version

^e Cronbach's alphas, parent version

Content analyses of the dimensions revealed that a three-factor model could reveal a better fit, namely "epilepsy-related physical complaints", "seizures" and "daily management". This model indicated a good fit, χ^2 was 36.46 ($N=89$, $Df=32$, $p=.269$), GFI was .93, AGFI was .88 and RMR was .05. The total coefficient of determination was .98. The PHI value between the first and second factor was .46, between the first and third factor .69 and between the second and third factor .64. Cronbach's alphas of these scales in the child version ranged from .70 to .78. Cronbach's alphas of these scales in the parent version ranged from .72 to .83. In Table 5.12 the results of confirmatory factor analysis and reliability testing are presented.

The LISREL analyses were conducted on the feelings items. The resulting dimensions were used to form the corresponding prevalence scales, so that the epilepsy-specific section consists of three dimensions each including two scales. The first dimension

includes a scale measuring the prevalence of epilepsy-related physical complaints and a scale measuring feelings towards these complaints (each of 3 items). The second dimension includes a scale measuring the prevalence of seizures and a scale measuring feelings towards these seizures (each of 3 items). The third dimension includes a scale measuring the prevalence of daily management and a scale measuring feelings towards daily management (each of 4 items). The epilepsy-specific section thus consists of 20 items.

5.15 DISEASE-SPECIFIC SECTION FOR CHILDREN WITH JUVENILE CHRONIC ARTHRITIS

Table 5.13: *Lambda values, corrected item-total correlations and Cronbach's alphas for the juvenile chronic arthritis-specific section of the HOW ARE YOU? (HAY)*

Scale Item	Lambda ^a	Item-total corr. ^b	Alpha ^c	Item-total corr. ^b	Alpha ^c
I. Physical complaints			.73		.81
Does it bother you when you					
- have trouble moving	.38	.43		.62	
- have swollen joints	.46	.54		.60	
- have painful joints	.54	.67		.71	
- are stiff in the morning due to your juvenile chronic arthritis	.42	.45		.59	
II. Limitations			.74		.73
Does it bother you when you					
- have trouble getting dressed	.41	.49		.50	
- need to get up earlier because you need more time due	.39	.41		.62	
- have trouble writing due	.69	.68		.46	
- have to rest during the day because	.40	.40		.48	
- have to cancel an appointment because	.43	.49		.40	
III. Daily management			.76		.74
Does it bother you when you					
- have to take medication for your juvenile chronic arthritis	.49	.46		.39	
- have to do exercises for your juvenile chronic arthritis	.64	.63		.72	
- have to have physical therapy	.54	.43		.33	
- have to wear (one or more) splints					

Note. The parents' own feelings are assessed in the parent version

^a All lambda values are significant at $p < .002$

^b Item-total correlations, child version

^c Cronbach's alphas, child version

^d Item-total correlations, parent version

^e Cronbach's alphas, parent version

As described above, it was hypothesized that the disease-specific section for children with juvenile chronic arthritis consisted of the factors, "juvenile chronic arthritis-related physical complaints" and "daily management". This model indicated however

an unsatisfactory fit. The two-factor model had a χ^2 of 152.33 ($N=53$, $Df=76$, $p=.000$), GFI was .75, AGFI was .65, and RMR was .08. The total coefficient of determination was .95. Two items had a squared multiple correlation lower than .2. Cronbach's alphas of the scales were respectively .78 and .77. Two items were deleted because of decreasing alpha.

Content analyses of the dimensions revealed that a three-factor model could reveal a better fit, namely "juvenile chronic arthritis-related physical complaints", "limitations caused by juvenile chronic arthritis" and "daily management". This model indicated a better fit, χ^2 was 99.42 ($N=53$, $Df=62$, $p=.002$), GFI was .83, AGFI was .75, and RMR was .07. The total coefficient of determination was .99. The PHI value between the first and second factor was .60, between the first and third factor .41 and between the second and third .37. Cronbach's alphas of these scales in the child version ranged from .70 to .78. Cronbach's alphas of these scales in the parent version ranged from .74 to .81. In Table 5.13 the results of confirmatory factor analysis and reliability testing are presented.

The LISREL analyses were conducted on the feelings items. The resulting dimensions were used to form the corresponding prevalence scales, so that the juvenile chronic arthritis-specific section consists of three dimensions, each including two scales. The first dimension includes the prevalence of juvenile chronic arthritis-related physical complaints and feelings towards these complaints (each of 4 items). The second dimension includes the prevalence of limitations and feelings towards these limitations (each of 5 items). The third dimension includes the prevalence of daily management and the feelings towards daily management (each of 4 items). The resulting juvenile chronic arthritis-specific section thus consists of 26 items.

5.16 CONCURRENCE BETWEEN THE HAY AND OTHER MEASURES

Table 5.14: Spearman Rank correlations between CATIS and scales of the HAY

	CATIS	Feelings of inferiority	Concerns
CATIS	1.00		
Feelings of inferiority	.53*	1.00	
Concerns	.59*	.52*	1.00

* $p < .01$ (two-tailed)

It was hypothesized that the dimensions of the HAY measuring "concerns about illness" and "feelings of inferiority because of illness", were correlated with the CATIS, indicating concurrence between both instruments. In Table 5.14 the Spearman Rank correlations between the scales are presented.

In addition, it was hypothesized that the CBCL scales measuring physical competence, school competence, social competence and physical complaints were measuring the same latent factors as the scales measuring the quality of performance on physical activities, cognitive tasks, social activities, and the prevalence of physical complaints of the HAY. A multitrait-multimethod analysis using LISREL 8 revealed however an improper solution, as it was difficult to identify the model with only two methods and four traits (Kiers, Takane & ten Berge, 1996). To assess the correlation between the CBCL and the HAY, Spearman Rank correlations were used (Table 5.15).

Table 5.15: Spearman Rank correlations between scales of the CBCL and scales of the parent version of the HAY

CBCL	HAY parent version	Spearman Rank correlation
Activity competence	Quality of performance on physical activities	.16*
School competence	Quality of performance on cognitive activities	.58*
Social competence	Quality of performance on social activities	.26*
Physical complaints	Physical complaints	-.47*

* $p < .01$ (two-tailed)

All correlations between the subscales of the CBCL and the HAY were significant. The correlation on cognitive competence was the highest, whereas the correlation on physical competence was relatively low.

5.17 DIFFERENCES BETWEEN CHILDREN WITH A CHRONIC ILLNESS AND HEALTHY CHILDREN

5.17.1 Transformations and homogeneity of variance

Before applying the ANOVA analyses, transformation was necessary for the scales measuring frequency of physical complaints, the quality of performance on physical activities, cognitive tasks and social activities of the child as well as the parent version ($X_{new} = 1/(5 - X_{old})$).

Levene's tests for homogeneity of variances of the scales in the *child version* were significant ($p < .01$) for the scales measuring the prevalence of physical activities, quality of performance on physical and social activities and the scales measuring feelings towards limitations in physical and social activities. In the scale measuring feelings towards physical limitations, the variance was highest in the smaller group, so that a significance level of the F-statistics of .001 was used for this scale (see paragraph 5.6).

Levene's tests for homogeneity of variances of the scales in the *parent version* were significant ($p < .01$) for the scales measuring the prevalence of physical complaints, quality of performance on physical activities, cognitive tasks and social activities and the scales measuring feelings towards limitations in social activities and physical complaints. In the scale measuring feelings towards physical complaints, the variance was highest in the smaller group, so that a significance level of the F-statistics of .001 was used for this scale.

5.17.2 Comparisons of means according to child report

Differences between the two groups were examined using General Linear Models (GLM) (SPSS) to perform a two-factorial analysis of variance (ANOVA) with gender and illness as independent factors and age as covariate. The analyses of variance were thus administered on the adjusted means for age. To be able to interpret the findings, the observed means and standard deviations of the child version are presented (see Table 5.16).

Table 5.16: Means and standard deviations of scales according to child report

	Ill children M (SD)	Healthy children M (SD)
Prevalence		
Physical activities	2.63 (.54)	2.60 (.49)
Cognitive tasks	3.14 (.58)	3.07 (.58)
Social activities	2.29 (.52)*	2.37 (.48)
Physical complaints	3.52 (.56)	3.57 (.51)
Happiness	3.12 (.63)	3.07 (.62)
Quality of performance		
Physical activities	3.76 (.41)*	3.91 (.18)
Cognitive tasks	3.75 (.38)	3.74 (.36)
Social activities	3.86 (.31)	3.90 (.23)
Feelings		
Physical activities	2.38 (.71)	2.52 (.75)
Cognitive tasks	2.21 (.76)	2.18 (.68)
Social activities	2.16 (.68)*	2.39 (.62)
Physical complaints	1.95 (.65)	1.90 (.64)

Note. Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations and a low level of negative feelings

* Significant effects for illness ($p < .01$)

As expected, the total group of girls reported a significantly lower prevalence of physical activities (boys: $M=2.72$, $SD=.48$; girls: $M=2.52$, $SD=.54$, $p < .01$), a higher prevalence of physical complaints (boys: $M=3.62$, $SD=.50$; girls: $M=3.47$, $SD=.57$, $p < .01$) and more negative feelings about having physical complaints (boys: $M=1.99$, $SD=.69$; girls: $M=1.87$, $SD=.59$, $p < .01$) than the total group of boys did. No significant interaction effects between having a chronic illness and gender were found.

Children with a chronic illness scored significantly lower than healthy children on the prevalence of social activities and the quality of performance on physical activities ($p < .01$). In addition, scores on feelings towards limitations in social activities were significantly lower ($p < .01$), revealing more negative feelings.

5.17.3 Comparisons of means according to parent report

Differences between the two groups were examined using General Linear Models (GLM) (SPSS) to perform a two-factorial analysis of variance (ANOVA) with gender and illness of the child as independent factors and child's age as covariate. The observed means and standard deviations of the parent version are presented (see Table 5.17).

As was also found in child reports, parents of the total group of girls reported a lower prevalence of physical activities (boys: $M=2.68$, $SD=.53$; girls: $M=2.49$, $SD=.55$) and a higher level of physical complaints for their children than parents of the total group of boys did (boys: $M=3.68$, $SD=.46$; girls: $M=3.61$, $SD=.49$, $p < .01$). No significant interaction effect between having a chronic illness and gender was found.

Table 5.17: Means and standard deviations of scales parent report

	Ill children M (SD)	Healthy children M (SD)
Prevalence		
Physical activities	2.57 (.52)	2.60 (.52)
Cognitive tasks	3.03 (.59)	3.08 (.56)
Social activities	2.25 (.45)*	2.38 (.48)
Physical complaints	3.58 (.53)*	3.75 (.38)
Happiness	2.94 (.54)	2.99 (.50)
Quality of performance		
Physical activities	3.73 (.46)*	3.96 (.14)
Cognitive tasks	3.69 (.52)*	3.82 (.31)
Social activities	3.77 (.43)*	3.91 (.27)
Feelings		
Physical activities	2.17 (.68)	2.24 (.58)
Cognitive tasks	1.90 (.62)	1.98 (.57)
Social activities	1.84 (.58)*	2.03 (.51)
Physical complaints	1.97 (.62)	2.09 (.66)

Note. Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations and a low level of negative feelings

* Significant effects for illness ($p < .01$)

According to parent report, children with a chronic illness had a significantly lower prevalence of social activities, more physical complaints, and a lower quality of performance on physical activities, cognitive tasks and social activities than healthy children ($p < .01$). In addition, parents of children with a chronic illness had more negative feelings towards limitations in social activities ($p < .01$).

5.18 DIFFERENCES BETWEEN CHILDREN WITH DISTINCT CHRONIC ILLNESSES

Table 5.18: Mean scores and standard deviations of distinct groups by child report

	Asthma M (SD)	IDDM M (SD)	Epilepsy M (SD)	JCA M (SD)	Healthy M (SD)
Prevalence					
Physical activities	2.57 (.56) ^d	2.78 (.49) ^{ha}	2.70 (.45)	2.48 (.65)	2.60 (.49)
Cognitive tasks	3.09 (.66)	3.23 (.57)	3.15 (.58)	3.20 (.65)	3.07 (.58)
Social activities	2.24 (.52) ^h	2.31 (.52)	2.36 (.49)	2.34 (.56)	2.37 (.48)
Physical complaints	3.47 (.58)	3.53 (.53)	3.60 (.55)	3.59 (.54)	3.57 (.52)
Happiness	3.06 (.63)	3.16 (.64)	3.10 (.63)	3.18 (.59)	3.07 (.62)
Concerns related to illness	3.74 (.42) ^d	3.58 (.47) ^{ae}	3.84 (.30) ^d	3.68 (.51)	
Feelings of inferiority	3.73 (.43)	3.81 (.31) ^j	3.84 (.28) ^j	3.60 (.43) ^{de}	
Quality of performance					
Physical activities	3.69 (.42) ^{hed}	3.91 (.23) ^{aj}	3.89 (.23) ^{aj}	3.47 (.61) ^{hed}	3.91 (.18)
Cognitive tasks	3.75 (.40)	3.79 (.39)	3.68 (.38)	3.82 (.25)	3.74 (.36)
Social activities	3.82 (.35) ^{he}	3.88 (.24)	3.92 (.24) ^a	3.88 (.28)	3.90 (.23)
Feelings					
Physical activities	2.44 (.70)	2.40 (.72)	2.31 (.73)	2.17 (.69) ^h	2.52 (.75)
Cognitive tasks	2.27 (.80)	2.21 (.71)	2.09 (.80)	2.15 (.59)	2.18 (.68)
Social activities	2.16 (.69) ^h	2.22 (.61)	2.17 (.73)	2.04 (.65) ^h	2.39 (.62)
Physical complaints	1.96 (.64)	1.86 (.65)	1.98 (.73)	2.06 (.52)	1.90 (.64)

Note. Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations and a low level of negative feelings

^h Mean significantly different from healthy

^a Mean significantly different from asthma

^d Mean significantly different from diabetes mellitus (IDDM)

^e Mean significantly different from epilepsy

^j Mean significantly different from juvenile chronic arthritis (JCA)

In the following paragraphs the results for the four disease-specific groups are presented separately. Differences between groups were established using General Linear Models (GLM) (SPSS) to perform a two-factorial analysis of variance (ANOVA) with gender and illness as independent factors and age as covariate. In Tables 5.18 and 5.19, the means and standard deviations of the distinct groups of respectively the child version and parent version are presented. A transformation was performed on the subscales prevalence of physical complaints and quality of performance on physical activities, cognitive tasks, social activities of the child as well as the parent version ($X_{new} = 1/(5 - X_{old})$). As the group of healthy children was only used as a reference group, the results of healthy children are not discussed in a separate paragraph.

Table 5.19: Mean scores and standard deviations of distinct groups by parent report

	Asthma M (SD)	IDDM M (SD)	Epilepsy M (SD)	JCA M (SD)	Healthy M (SD)
Prevalence					
Physical activities	2.51 (.57) ^d	2.74 (.56) ^{haj}	2.68 (.49)	2.29 (.62) ^d	2.60 (.52)
Cognitive tasks	2.98 (.62)	3.15 (.50)	2.97 (.61)	3.05 (.58)	3.08 (.56)
Social activities	2.22 (.44) ^{hd}	2.35 (.43) ^a	2.23 (.46)	2.19 (.47)	2.38 (.48)
Physical complaints	3.54 (.58) ^h	3.61 (.43) ^h	3.63 (.49)	3.57 (.53)	3.75 (.38)
Happiness	2.95 (.54)	2.98 (.55)	2.93 (.53)	2.86 (.55)	2.99 (.50)
Concerns related to illness	3.61 (.51) ^{jd}	3.16 (.57) ^{ae}	3.55 (.56) ^{jd}	3.26 (.64) ^{ae}	
Feelings of inferiority	3.75 (.36) ^{ji}	3.75 (.34)	3.78 (.38) ^{ji}	3.53 (.54) ^{ae}	
Quality of performance					
Physical activities	3.67 (.44) ^{hde}	3.90 (.36) ^{aj}	3.86 (.26) ^{aj}	3.39 (.69) ^{hde}	3.96 (.14)
Cognitive tasks	3.69 (.51)	3.76 (.49)	3.53 (.63) ^{hj}	3.81 (.37) ^e	3.82 (.31)
Social activities	3.72 (.43) ^h	3.83 (.44)	3.86 (.36)	3.73 (.47)	3.91 (.27)
Feelings					
Physical activities	2.27 (.69) ^{ji}	2.08 (.66)	2.18 (.73)	1.93 (.51) ^{ha}	2.24 (.58)
Cognitive tasks	1.94 (.64)	1.86 (.57)	1.86 (.66)	1.92 (.56)	1.98 (.57)
Social activities	1.89 (.61)	1.79 (.58)	1.81 (.57)	1.77 (.48) ^h	2.03 (.51)
Physical complaints	2.06 (.64)	1.89 (.60)	1.91 (.60)	1.85 (.57)	2.09 (.66)

Note. Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations and a low level of negative feelings

^h Mean significantly different from healthy

^a Mean significantly different from asthma

^d Mean significantly different from diabetes mellitus (IDDM)

^e Mean significantly different from epilepsy

^j Mean significantly different from juvenile chronic arthritis (JCA)

5.18.1 Comparison of children with asthma with other groups.

As was hypothesized, children with asthma, according to *child report*, scored significantly lower on the prevalence of social activities ($p < .01$), lower on the quality of performance of physical activities ($p < .001$) and lower on the quality of performance on social activities than healthy children ($p < .001$). In addition, children with asthma scored significantly more negative feelings towards limitations in social activities than healthy children did ($p < .01$). Although children with asthma had the lowest score on physical complaints, implying more physical complaints, they did not report significantly more physical complaints than healthy children in contrast to expectations, nor did they report a lower prevalence of cognitive tasks.

In respect of differences between disease-specific groups, children with asthma scored significantly lower on the prevalence of physical activities than children with diabetes mellitus ($p < .01$), lower on the quality of performance of physical activities than children with epilepsy and diabetes mellitus ($p < .01$) and lower on the quality of performance on social activities than children with epilepsy ($p < .01$). Furthermore, children with asthma scored fewer concerns related to their illness than children with diabetes mellitus ($p < .01$).

As was hypothesized, according to *parent report*, children with asthma scored significantly lower on the prevalence of social activities ($p < .01$), significantly more physical complaints than healthy children ($p < .01$), and lower on the quality of performance on physical activities and social activities than healthy children ($p < .001$). In contrast to what was expected, parents of children with asthma didn't report a lower prevalence of cognitive tasks.

In addition, according to their parents, children with asthma scored significantly lower on the prevalence of physical activities and on the prevalence of social activities than children with diabetes mellitus ($p < .01$), lower on the quality of performance of physical activities than children with diabetes mellitus and epilepsy ($p < .01$). In contrast, parents of children with asthma scored significantly fewer negative feelings about limitations in physical activities than parents of children with juvenile chronic arthritis did ($p < .01$). Furthermore, parents of children with asthma reported fewer concerns about their child's illness than parents of children with juvenile chronic arthritis and diabetes mellitus ($p < .01$) did and fewer feelings of inferiority than parents of children with juvenile chronic arthritis did ($p < .01$).

5.18.2 Comparison of children with diabetes mellitus with other groups

In contrast to what was expected, according to *child report*, no significant differences between children with diabetes mellitus and healthy children were found on social activities. In addition, children with diabetes mellitus scored a significantly higher prevalence of physical activities than healthy children ($p < .01$).

With respect to differences between disease-specific groups, children with diabetes mellitus scored significantly higher on the prevalence of physical activities than children with asthma and higher on the quality of performance on physical activities than children with asthma ($p < .01$) and children with juvenile chronic arthritis ($p < .001$). In contrast, children with diabetes mellitus scored significantly more concerns about their illness than children with asthma and children with epilepsy ($p < .01$) but significantly fewer feelings of inferiority than children with juvenile chronic arthritis ($p < .01$).

In contrast to what was expected, according to *parent report*, children with diabetes mellitus scored significantly higher on the prevalence of physical activities than healthy children, indicating a higher prevalence of physical activities. In addition, parents reported more physical complaints of their children than parents of healthy children did ($p < .01$). In contrast to what was expected, according to *parent report*, no significant differences were found on social activities

With respect to differences between groups, according to their parents, children with diabetes mellitus scored significantly higher on the prevalence of physical

activities than children with asthma and juvenile chronic arthritis ($p < .01$), significantly higher on the prevalence of social activities than children with asthma ($p < .01$), significantly higher on the quality of performance on physical activities than children with asthma ($p < .01$) and juvenile chronic arthritis ($p < .001$), revealing a better level of functioning. In contrast, parents of children with diabetes mellitus reported significantly more concerns about their child's illness than parents of children with asthma and epilepsy did ($p < .01$).

5.18.3 Comparison of children with epilepsy with other groups

In contrast to what was expected, according to the *child report*, there were no significant differences between children with epilepsy and healthy children on cognitive tasks or social activities.

Compared to other disease-specific groups, children with epilepsy scored significantly higher on the quality of performance on physical activities than children with juvenile chronic arthritis ($p < .001$) and children with asthma ($p < .01$) and significantly higher on the quality of performance on social activities than children with asthma ($p < .001$). In addition, children with epilepsy reported significantly fewer concerns about their illness than children with diabetes mellitus and less feelings of inferiority than children with juvenile chronic arthritis ($p < .01$).

According to *parent report*, as was hypothesized, children with epilepsy scored significantly lower on the quality of performance on cognitive tasks than healthy children ($p < .001$). No significant results were, however, found on social activities. Furthermore, parents scored a significantly higher quality of performance of their children on physical activities than parents of children with asthma and juvenile chronic arthritis did ($p < .001$) and a lower quality of performance on cognitive tasks than parents of children with juvenile chronic arthritis did ($p < .01$). In addition, parents of children with epilepsy scored significantly fewer concerns about their child's illness than parents of children with diabetes mellitus or juvenile chronic arthritis did ($p < .01$) and fewer feelings of inferiority related to their child's illness than parents of children with juvenile chronic arthritis did ($p < .01$).

5.18.4 Comparison of children with juvenile chronic arthritis with other groups

As was hypothesized, according to *child report*, children with juvenile chronic arthritis scored significantly lower on the quality of performance on physical activities than healthy children. In addition, children with juvenile chronic arthritis had more negative feelings related to limitations in physical and social activities ($p < .01$) than healthy children. In contrast to what was expected, no significant differences were found on the prevalence of cognitive tasks the prevalence of or quality of performance on social activities or the prevalence of physical complaints.

Compared to other disease-specific groups, children with juvenile chronic arthritis scored significantly lower on the quality of performance on physical activities than children with epilepsy or diabetes mellitus ($p < .001$). Furthermore, children with juvenile chronic arthritis reported significantly more feelings of inferiority related to their illness than children with diabetes mellitus and epilepsy did ($p < .01$).

As was hypothesized, according to *parent report*, children with juvenile chronic arthritis scored significantly lower on the quality of performance on physical activities than healthy children ($p < .001$), but not significantly lower on the prevalence of physical activities. In addition, parents had more negative feelings towards limitations in physical activities ($p < .001$) and social activities ($p < .01$) than parents of healthy children. In contrast to expectations, no significant differences were found on the prevalence of cognitive tasks, the prevalence of or quality of performance on social activities or the prevalence of physical complaints.

Compared to other disease-specific groups, according to parent report, children with juvenile chronic arthritis, scored significantly lower on the prevalence of physical activities than children with diabetes mellitus ($p < .001$), significantly lower on the quality of performance on physical activities than children with diabetes mellitus or epilepsy ($p < .001$), and significantly higher on the quality of performance on cognitive tasks than children with epilepsy ($p < .01$). Furthermore, parents of children with juvenile chronic arthritis had significantly more negative feelings about limitations in physical activities than parents of children with asthma ($p < .01$). In addition, parents had more concerns and more feelings of inferiority related to their child's illness than parents of children with asthma and epilepsy had ($p < .001$).

5.19 RELATION BETWEEN CURRENT DISEASE ACTIVITY AND CHILD'S FUNCTIONING

It was hypothesized that within a specific diagnostic group more symptoms or a greater severity of symptoms correlates with more general physical complaints, and more limitations in physical, social activities, and in cognitive tasks.

Parents were asked to rate the number of symptoms and the severity of their child's illness within each disease-specific group, which was called "current disease activity". Within each of the disease-specific groups, the correlation was assessed between current disease activity and physical complaints and limitations in physical and social activities and in cognitive tasks. Multiple hierarchical regression analyses were performed to test the correlation of disease activity with the scales of the HAY of child report and parent report after controlling for child's age and gender. Transformation was performed for the scales measuring prevalence of physical complaints, the quality of performance on physical activities, cognitive tasks and social activities of the child as well as the parent version ($X_{\text{new}} = 1/(5 - X_{\text{old}})$).

5.19.1 Relation between disease activity and functioning in children with asthma

For children with asthma, parents rated the amount of coughing during the day or night, breathlessness and wheezing. All items were dichotomized, with 0 having no asthma-related symptoms and 1 having asthma-related symptoms. The parent-reported asthma severity scale ranged thus from 0-3, with 0 having no symptoms at all and 3 having symptoms on all variables. In this way, 124 children (45%) had no asthma-related symptoms at all, 74 (27%) had 1 asthma-related symptom, 56 (20%) had 2 asthma-related symptoms and 18 (7%) had 3 asthma-related symptoms.

A significant standardized regression coefficient between current disease activity and *child-reported functioning* was found for the prevalence of physical complaints ($\beta=-.22$; $p<.001$), the quality of performance on physical activities ($\beta=-.27$; $p<.001$) and the quality of performance on social activities ($\beta=-.22$; $p<.001$). A significant standardized regression coefficient between current disease activity and *parent-reported functioning* was found for the prevalence of physical complaints ($\beta=-.18$; $p<.01$), the quality of performance on physical activities ($\beta=-.35$; $p<.001$) and the quality of performance on social activities ($\beta=-.17$; $p<.01$).

5.19.2 Relation between disease activity and functioning in children with diabetes mellitus

For children with diabetes mellitus, parents rated how often their children experienced a hypoglycemia and a hyperglycemia during the past seven days in the daytime and a hypoglycemia at night. All items were dichotomized, with 0 having no diabetes mellitus-related symptoms and 1 having diabetes mellitus-related symptoms. The parent-reported diabetes mellitus activity scale ranged thus from 0-3, with 0 having no diabetes mellitus-related symptoms at all and 3 having diabetes mellitus-related symptoms on all variables. In this way, 31 children (22%) had no diabetes mellitus-related symptoms at all, 50 (36%) had 1 diabetes mellitus-related complaint, 42 (30%) had 2 diabetes mellitus-related symptoms, 16 (12%) had 3 diabetes mellitus-related symptoms.

A significant standardized regression coefficient between current disease activity and *child-reported functioning* was found for the prevalence of physical activities ($\beta=-.24$; $p<.01$). No significant standardized regression coefficients between current disease activity and *parent-reported functioning* were found.

5.19.3 Relation between disease activity and functioning in children with epilepsy

For children with epilepsy, parents rated whether there were any epilepsy symptoms during the past six months and how serious these symptoms were. The items were dichotomized, with 0 having no epilepsy-related symptoms and 1 having epilepsy related symptoms. The parent-reported epilepsy activity scale ranged thus from 0 to 2, with 0 having no symptoms at all and 2 having serious symptoms according to their parents. In this way, 41 children (39%) had no epilepsy-related symptoms, 42

(40%) had no serious epilepsy-related symptoms, 21 (20%) had serious epilepsy-related symptoms.

No significant standardized regression coefficients were found between current disease activity and child-reported physical or social activities or cognitive tasks.

A significant standardized regression coefficient between current disease activity and *parent-reported functioning* was found for the quality of performance on physical activities ($\beta = -.27$; $p < .01$).

5.19.4 Relation between disease activity and functioning in children with juvenile chronic arthritis

For children with juvenile chronic arthritis, parents specified which joints had troubled the child during the past half year and which symptoms related to the juvenile chronic arthritis their child had experienced during the past half year. All items were dichotomized, with 0 having no juvenile chronic arthritis-related symptoms and 1 having juvenile chronic arthritis-related symptoms. The parent-reported disease activity scale ranged thus from 0-16, with 0 having no symptoms at all and 16 having symptoms on all variables. Six categories were made with 6 children (10%) having no juvenile chronic arthritis-related symptoms, 11 children (19%) having 1 to 3 juvenile chronic arthritis-related symptoms, 17 children (29%) having 4 to 6 juvenile chronic arthritis-related symptoms, 10 children (17%) having 7 to 9 juvenile chronic arthritis-related symptoms, 9 children (15%) having 10 to 12 juvenile chronic arthritis-related symptoms, 6 children (10%) having 13 or more juvenile chronic arthritis-related symptoms.

A significant standardized regression coefficient between current disease activity and *child-reported functioning* was found for the prevalence of physical activities ($\beta = -.59$; $p < .0012$), the prevalence of social activities ($\beta = -.44$; $p < .001$), the prevalence of physical complaints ($\beta = -.40$; $p < .01$), the quality of performance on physical activities ($\beta = -.53$; $p < .001$) and the quality of performance on social activities ($\beta = -.37$; $p < .01$). A significant standardized regression coefficient between current disease activity and *parent-reported functioning* was found for the prevalence of physical activities ($\beta = -.55$; $p < .001$), the prevalence of physical complaints ($\beta = -.42$; $p < .001$), the quality of performance on physical activities ($\beta = -.63$; $p < .001$) and the quality of performance on social activities ($\beta = -.44$; $p < .001$).

5.20 RELATION BETWEEN CHILD REPORT AND PARENT REPORT

To assess the concurrence between children and parents, intra-class correlations (ICC) were calculated between the children and parents of the group of children with a chronic illness and healthy children separately. For the scales measuring

prevalence of physical complaints and quality of performance on physical activities, cognitive tasks, social activities of the child as well as the parent version, the transformed scores were used ($X_{\text{new}} = 1/(5 - X_{\text{old}})$). In Table 5.20, the ICCs are presented for both groups.

Table 5.20: *Intraclass correlations between children and parents*

Scales	Children with a chronic illness	Healthy children
	ICC	ICC
Prevalence		
Physical activities	.60	.59
Cognitive tasks	.37	.37
Social activities	.49	.52
Physical complaints	.61	.45
Happiness	.36	.32
Quality of performance		
Physical activities	.65	.30
Cognitive tasks	.51	.35
Social activities	.44	.27

In the group of healthy children, higher ICCs were found on the prevalence of physical and social activities scales than on the scales measuring quality of performance on physical activities and social activities. In the group of children with a chronic illness, this difference was not found. In the group of children with a chronic illness, the concurrence between parents and children was higher on the quality of performance scale compared to the ICCs found between healthy children and their parents on these scales.

The ICCs were fairly comparable with Pearson or Spearman Rank correlations (Appendix Table 5.21). Student t tests revealed (Appendix Table 5.22) that in the group of children with a chronic illness, parents and children differed significantly on the prevalence of physical activities and cognitive tasks, prevalence of physical complaints, the child's happy feelings and the quality of performance on cognitive tasks and social activities ($p < .01$). In this group, parents scored a significantly lower prevalence of physical activities, cognitive tasks, and less happy feelings of their child, as well as a lower quality of performance on cognitive tasks and social activities than their children did ($p < .01$). In contrast, the parents scored fewer physical complaints than their children did ($p < .01$).

In the healthy group, children and parents differed significantly on the prevalence of physical complaints and the quality of performance on physical activities and cognitive tasks ($p < .01$) with parents scoring fewer physical complaints than their children did. In contrast to the group of children with a chronic illness, in the healthy group of children, parents scored a better quality of performance on

cognitive tasks than their children did ($p < .01$). Scales assessing feelings towards limitations or complaints were not compared using ICC because of distinct items measuring the parents' respectively the child's own feelings.

5.21 CONCLUSION AND DISCUSSION

5.21.1 Sample

A large group of children and their parents cooperated in this stage of the study, so that the validity and reliability testing of the HAY could be performed properly. Comparison of the demographic characteristics of the sample with prevalence estimates described in Chapter 1, revealed that the numbers of children in each group and the gender distributions in the distinct illness groups more or less reflect the prevalence of these illnesses in the general population. As expected, the group of children with asthma was the largest group involved. The group of children with juvenile chronic arthritis was the smallest group. As in the general population, the group of children with asthma included more boys than girls whereas the group of children with juvenile chronic arthritis included more girls than boys. The relative numbers of children with asthma as well as children with juvenile chronic arthritis meeting severe or considerable limitations according to parents were relatively large compared to the other groups, as was also described in the large NHIS study held in the US (Newacheck & Taylor, 1992). Children with a chronic illness were mainly recruited in hospitals, although a relatively large proportion of children with asthma were recruited through their GP. This is in accordance with the general population estimates. Healthy children were recruited through their schools.

Although the group of children with a chronic illness included was relatively large in comparison to other studies and seems to be similar to general prevalence estimates, the group of children with juvenile chronic arthritis was small. In addition, neither the type of juvenile chronic arthritis nor the type of seizures in epilepsy were involved in analyses as physician's reports on these characteristics were not available and group sizes would become very small. Furthermore, the illness duration or severity were not taken into account, as these are also dependent on the diagnosis. In addition, it must be noted that only children with no other major chronic illnesses and children who could fluently read and write in order to complete the questionnaire, were included. Nonresponse was not calculated, but data revealed that for the centers and schools for which nonresponse could be calculated, acceptable response percentages from 70% to 80% occurred. A bias in the results presented as well as indefinite results may, however, occur.

Children younger than 7 years and older than 13 years were deleted from analyses, as it was reported that the content as well as the layout of the questionnaire had to

be age-specific (Starfield, 1987; Stone & Lemanek, 1990; Flanery, 1990). For these age groups, new versions of the HAY should be developed.

5.21.2 Utility

As the children and parents filled in their questionnaire at home, parents were asked to indicate whether the child had filled in the questionnaire alone. According to the parents, most children were able to answer the HAY without help. Whether there was no influence of parents on their children answers, couldn't however be checked. Correlations between parents and children could be influenced by this factor as well as the possibility that children might have filled in their questionnaire rather too optimistically so as not to worry their parents. Comparable correlations between means were however noted in earlier research (Achenbach, McConaughy & Howell, 1987; Fritz & Overholser, 1989; Tyc, Mulhern, Fairclough, Ward, Relling & Longmore, 1993) indicating that both reports were not identical and should be used complementary.

5.21.3 Confirmatory factor analysis and reliability testing

Confirmatory Factor Analyses revealed a good fit and good reliabilities for the generic section of the HAY, consisting of five dimensions, measuring physical activities, cognitive tasks and social activities, physical complaints and happiness. Three dimensions were however deleted, including social problems, general treatment characteristics and negative emotional feelings. Although a reasonably good fit and reliabilities could be obtained by including social problems, it was considered that a better model and reasons of parsimony should predominate. Unfortunately, as a consequence, an assessment on this important area of functioning is missing. The reliability of the dimensions general treatment and negative emotions were too low. Reasons of low reliabilities and parsimony led to the decision to delete these two dimensions as well. It was believed that the assessment of these aspects in the chronic illness section, where they are also included, probably leads to more relevant items for the diagnostic groups involved.

The chronic illness section is applicable to all children with a chronic illness and consists of two dimensions with good reliabilities: the prevalence of concerns related to having a specific illness and the prevalence of feelings of inferiority because of having a specific illness.

As hypothesized, the asthma-specific section consists of two dimensions called "physical complaints" and "daily management" and was found to have a good fit and good reliabilities. In contrast to what was expected, the sections for children with epilepsy, diabetes mellitus and juvenile chronic arthritis each consists of three dimensions. Results revealed that the diabetes mellitus-specific section consists of three dimensions, including "physical complaints related to a hypo", "physical complaints related to a hyper" and "daily management" and was found to have a

good fit but low reliabilities. Although the reliabilities were considered to be too low, this model was nevertheless accepted. Further research is however needed in this group with reformulation and/or addition of items. The epilepsy-specific section also consists of three dimensions, including “physical complaints”, “seizures” and “daily management” with moderately good fit and good reliabilities. The juvenile chronic arthritis-specific section consists of three dimensions, including “physical complaints”, “limitations” and “daily management”, which was found to have only an acceptable fit but good reliabilities. Probably as a result of the small sample size the results of confirmatory factor analysis were not stable in this group (Saris & Stronkhorst, 1984; Jöreskog & Sörbom, 1989; Evans, 1994). New analyses using larger groups have to confirm the structure found in this group.

In general, parent reliabilities were somewhat higher than reliabilities found for scales in the child version.

5.21.4 Concurrence between the HAY and the CBCL and the CATIS

Correlations between scales measuring physical, cognitive and social tasks and activities and physical complaints by the CBCL and the quality of performance scale and physical complaints scale of the parent version of the HAY revealed that correlations on cognitive competence were fairly high, which indicates concurrence between these two scales. Although significant correlations were found between the physical and social dimensions of the HAY and the CBCL, correlations were low. An explanation for this discrepancy may be that the CBCL not only includes quality of performance but also the involvement of children in physical and social activities in the physical and social dimension, whereas the quality of performance scales of the HAY measure only competence. The moderate correlation on physical complaints may be explained by the fact that the CBCL measures physical complaints without a known physical cause when parents fill in the items properly. The concurrence between the CBCL and HAY supports the validity of the HAY as no high correlations could be expected when gold standards don't exist (Hillers et al., 1994).

The dimensions “prevalence of concerns about the illness” and “prevalence of feelings of inferiority because of having an illness” correlated significantly with the translated version of the CATIS (Austin & Huberty, 1993) for which good validity and reliability in this Dutch sample was found. The correlations between the child version of the HAY scales measuring “concerns” and “feelings of inferiority” and the CATIS were moderate and significant and confirm the validity of the HAY.

5.21.5 Differences between children with a chronic illness and healthy children.

Significant differences in the expected direction, according to child report, were found for the prevalence of social activities and the quality of performance on physical activities. In addition, children with a chronic illness scored more negative feelings on limitations in social activities than healthy children did. Contrary to our

expectations, children with a chronic illness did not report more general physical complaints compared to healthy children. An explanation for this finding might be that children with a chronic illness are more used to general physical complaints and thus ignore them more often. This finding stresses the importance of the assessment of disease-specific complaints because scales measuring general complaints will probably not detect fully the relevant physical symptoms children experience in daily life. Nor were there any significant differences found for the prevalence of physical activities or cognitive tasks or the quality of performance on cognitive tasks or social activities or feelings of happiness. Whether children underreport problems or limitations is not known.

Significant differences in the expected direction in the parent version were found for the prevalence of social activities, general physical complaints and the quality of performance on physical activities, cognitive tasks and social activities. Like their children, parents of children with a chronic illness had more negative feelings about limitations in social activities than parents of healthy children had.

No significant results were, however, found on the prevalence of physical activities and cognitive tasks. These results might be explained by the fact that, in general, rather than the prevalence of physical activities, the quality of performance in physical activities is affected for children with a chronic illness like the illnesses included in the study.

In addition, it was hypothesized that as school absenteeism is higher among children with a chronic illness, the prevalence of cognitive tasks would be lower than among healthy children. In contrast to what was expected, there were no significant results found in this direction, possibly as a consequence of the fact that this scale does not measure directly school absenteeism, such as the number of days lost at school during the previous week, but the prevalence of school related tasks. When children only missed one or two days during the previous week, the rated prevalence of cognitive tasks will probably not be influenced. Furthermore, no significant differences were found for the prevalence of happy feelings of the children.

5.21.6 Differences between boys and girls

As hypothesized, girls (as well as their parents) reported a lower prevalence of physical activities and a higher prevalence of physical complaints than boys did. The girls themselves reported also more negative feelings about physical complaints than boys. The fact that girls reported more physical symptoms than boys did, is in concordance with the general knowledge that women report more physical symptoms than men do (Mackenbach, 1994) and has been reported more often in studies among children (Hambley, Brazil, Farrow & Chua, 1989). No significant interaction effects were found for illness and gender, which indicates that main effects were

independent and consistent across groups. Distinct norms for boys and girls should therefore be developed.

5.21.7 Differences between children with asthma, IDDM, epilepsy and JCA and healthy children

It was hypothesized that due to disease-specific characteristics of illness, differences in the profiles of functioning of children in the diagnostic groups studied would occur. In order to establish the discriminative power of the HAY, differences between groups were established using General Linear Models (GLM) (SPSS) to perform a two-factorial analysis of variance (ANOVA) with gender and illness as independent factors and age as covariate. Tables 5.23 to 5.26 show which scales significant differences were found on for each disease-specific group.

Table 5.23: Differences between children with asthma and healthy children, confirmation of hypotheses

Dimensions	Scales	Child report	Parent report
Physical functioning	Prevalence of physical activities		
	Quality of performance on physical activities	*	*
	Prevalence of physical complaints		*
	Feelings towards limitations in physical activities		
	Feelings towards physical complaints		
Cognitive functioning	Prevalence of cognitive tasks		
	Quality of performance on cognitive tasks		
	Feelings towards limitations in cognitive tasks		
Social functioning	Prevalence of social activities	*	*
	Quality of performance on social activities	*	*
	Feelings towards limitations in social activities	*	
Happiness	Prevalence of happy feelings		

Note. * Indicates that a significant result on the scale was found. For children with asthma, it was hypothesized that they would experience more physical complaints and more limitations in physical activities as well as in social activities and would have more school absenteeism than healthy children

As hypothesized, *children with asthma* scored a lower quality of performance on physical activities than healthy children according to child and parent report. As hypothesized, children with asthma also had more physical complaints than healthy children according to their parents. In addition, parents and children scored a lower prevalence of social activities and a lower quality of performance on social activities than healthy children as was hypothesized. As the social activities scale of the HAY measures participation in activities with other children that are often related to physical activities in this age group, this result is in line with the review of studies presented in Chapter 2. For instance, playing with friends after school might be affected by your illness when you are not able to run or bike, to play football or rope-skipping. Furthermore, children with asthma also have more negative feelings

towards limitations in social activities than healthy children. Social activities should therefore be considered when studying or treating children with asthma.

In respect of differences between disease-specific groups, children with asthma scored a lower prevalence of physical activities than children with diabetes mellitus, according to child and parent report. In addition, children with asthma had a lower prevalence of social activities than children with diabetes mellitus according to parent report. Furthermore, children with asthma had a lower quality of performance on physical activities than children with diabetes mellitus and epilepsy, according to child and parent report. According to child report, they had a lower quality of performance on social activities than children with epilepsy. Whilst a lower performance in social functioning was expected in children with epilepsy, children with asthma might score lower because physical activities are often involved in this age group when playing with other children. Children with asthma had fewer concerns about their illness than children with diabetes mellitus. Parents of children with asthma had fewer concerns about their child's illness than parents of children with juvenile chronic arthritis and diabetes mellitus. Parents also had fewer negative feelings about limitations in physical activities than parents of children with juvenile chronic arthritis.

Table 5.24: Differences between children with diabetes mellitus and healthy children

Dimensions	Scales	Child report	Parent report
Physical functioning	Prevalence of physical activities	*	•
	Quality of performance on physical activities		
	Prevalence of physical complaints		•
	Feelings towards limitations in physical activities		
	Feelings towards physical complaints		
Cognitive functioning	Prevalence of cognitive tasks		
	Quality of performance on cognitive tasks		
	Feelings towards limitations in cognitive tasks		
Social functioning	Prevalence of social activities		
	Quality of performance on social activities		
	Feelings towards limitations in social activities		
Happiness	Prevalence of happy feelings		

Note. * Indicates that a significant result on the scale was found. For children with diabetes mellitus, it was hypothesized that they would experience more limitations in social functioning than healthy children

In contrast to what was expected, *children with diabetes mellitus* do not have more limitations in social activities than healthy children. Further research on social activities among the children with diabetes mellitus is needed, given the small number of studies on which the hypothesis concerning social functioning was based. In accordance with the results of the review of clinical studies among children with diabetes mellitus, children and/or their parents scored within average range or even

better on most other scales than healthy children or children with another diagnosis. On the physical activities scale, children with diabetes mellitus as well as their parents even scored significantly higher than healthy children or their parents did. Parents, however, reported more physical complaints of their children than parents of healthy children did. The illness characteristics of diabetes mellitus might explain the higher prevalence of complaints such as headaches or feeling nauseous, as these complaints can occur when hyperglycemia or hypoglycemia occur. Whether parents overreport these complaints or children underreport is however not known. In clinical practice these results may lead to a further investigations of the complaints, the attentiveness of parents and children towards illness signs as well as the feelings towards the illness.

Both children with diabetes mellitus and their parents reported more concerns about the illness than children with asthma or epilepsy and their parents did. This may be explained by the fact that short-term as well as long-term consequences of diabetes mellitus are apparent and is in line with the results of Simell, Moren, Keltokangas-Jarvinen, Hakalax and Simell (1995), who reported that two-thirds of the families had disease-specific fears. In addition, children with diabetes mellitus scored fewer feelings of inferiority than children with juvenile chronic arthritis did.

Table 5.25: Differences between children with epilepsy and healthy children

Dimensions	Scales	Child Report	Parent report
Physical functioning	Prevalence of physical activities		
	Quality of performance on physical activities		
	Prevalence of physical complaints		
	Feelings towards limitations in physical activities		
	Feelings towards physical complaints		
Cognitive functioning	Prevalence of cognitive tasks		
	Quality of performance on cognitive tasks		*
	Feelings towards limitations in cognitive tasks		
Social functioning	Prevalence of social activities		
	Quality of performance on social activities		
	Feelings towards limitations in social activities		
Happiness	Prevalence of happy feelings		

Note. * Indicates that a significant result on the scale was found. For children with epilepsy, it was hypothesized that they would experience more limitations on cognitive tasks and in social functioning than healthy children

The hypotheses with regard to the functioning of *children with epilepsy* were partially confirmed. As hypothesized, parents scored a lower quality of performance on cognitive tasks than healthy children, but their children didn't. Whether children underreport problems or parents are biased in their assessment because of their expectations (Aldenkamp et al., 1993; Suurmeyer, 1991) is not known. Parents nor

their children scored more limitations in social activities than healthy children. Again it must be stated that the hypothesis on social functioning was based on a limited number of studies and further research on social activities and problems among the children with epilepsy is needed.

According to parent report, children with epilepsy had a lower quality of performance on cognitive tasks than children with juvenile chronic arthritis. In addition, children with epilepsy had a higher quality of performance on physical activities than children with asthma or juvenile chronic arthritis according to the children as well as their parents, which might be explained by the illness-specific characteristics. Children themselves reported a higher quality of performance on social activities than children with asthma. Furthermore, they reported fewer concerns about their illness than children with diabetes mellitus and fewer feelings of inferiority than children with juvenile chronic arthritis. In addition, parents are less concerned about their child's illness than parents of children with diabetes mellitus and juvenile chronic arthritis and experience less feelings of inferiority than parents of children with juvenile chronic arthritis.

Table 5.26: Differences between children with juvenile chronic arthritis and healthy children

Dimensions	Scales	Child Report	Parent report
Physical functioning	Prevalence of physical activities		
	Quality of performance on physical activities	*	*
	Prevalence of physical complaints		
	Feelings towards limitations in physical activities	*	*
	Feelings towards physical complaints		
Cognitive functioning	Prevalence of cognitive tasks		
	Quality of performance on cognitive tasks		
	Feelings towards limitations in cognitive tasks		
Social functioning	Prevalence of social activities		
	Quality of performance on social activities		
	Feelings towards limitations in social activities	*	*
Happiness	Prevalence of happy feelings		

Note. * Indicates that a significant result on the scale was found. For children with juvenile chronic arthritis, it was hypothesized that they would experience more physical complaints, more limitations in physical and social activities and would have more school absenteeism than healthy children

As hypothesized, *children with juvenile chronic arthritis* and their parents scored a significantly lower quality of performance in physical activities than healthy children. In addition, both the children as well as their parents have more negative feelings towards limitations in physical activities than healthy children or their parents had. In addition, both parents and children scored more negative feelings towards limitations in social functioning than healthy children and their parents did. Although children with juvenile chronic arthritis also scored a lower prevalence of

physical activities according to child as well as parent report than healthy children, this difference was not significant. Nor were there any significant differences found in the prevalence of physical complaints or the prevalence and quality of performance in social activities. Most studies on which the hypotheses were based, however, included disease-specific physical complaints and it is in this area that problems are apparent.

The fact that no results were found on social activities is not easy to explain. Further study should include other variables, such as duration, severity or lower self concept, which proved to be important in predicting problems in social functioning (Daltroy, Larson, Eaton & Partridge, 1992; Ennett, DeVellis, Earp & Kredich, 1991; Ungerer, Horgen, Chaitow & Champion, 1988).

The illness-specific characteristics of juvenile chronic arthritis could explain why children with juvenile chronic arthritis, as well as their parents, report a lower quality of performance in physical activities than children with epilepsy or diabetes mellitus. Parents scored also a lower prevalence of physical activities of their child than parents of children with diabetes did. Parents scored a better quality of performance on cognitive tasks than parents of children with epilepsy did. Whereas both the parents of children with asthma and those with juvenile chronic arthritis report limitations in physical activities of their children, parents of children with juvenile chronic arthritis report significantly more negative feelings towards limitations in physical activities. This indicates that this area is important with respect to the treatment of children and the counseling of parents of children with juvenile chronic arthritis.

In addition, children with juvenile chronic arthritis had more feelings of inferiority than children with diabetes mellitus or epilepsy. Their parents reported more feelings of inferiority and more concerns related to their child's illness than parents of children with epilepsy or asthma did.

5.21.8 Sensitivity of the HAY for severity of symptoms

The sensitivity of the HAY for severity of symptoms, called current disease activity, within the distinct groups was assessed by relating symptoms as reported by the parents with the level of functioning of the child after controlling for age and gender.

Results revealed that more than half of the *children with asthma* had had asthma-specific symptoms during the previous seven days. The current disease activity appeared to correlate with the child-reported prevalence of physical complaints, and the quality of performance on physical and social activities. In addition, current disease activity correlated with the parent-reported prevalence of the child's physical complaints, and the quality of performance on physical and social activities

More than three quarters of the *children with diabetes mellitus* had had hyperglycemias or hypoglycemias during the past seven days. Whilst the disease activity correlated with the prevalence of physical activities according to child report, they did not correlate with parent-reported levels of functioning or complaints.

More than half of the group of *children with epilepsy* experienced (serious) epilepsy symptoms during the past six months. No significant relations between current disease activity and child reported functioning were found. Current disease activity correlated with the parent-reported quality of performance on physical activities.

Only 10% of the *children with juvenile chronic arthritis* had no current disease activity reported by the parents. For children with juvenile chronic arthritis, current disease activity correlated with the prevalence of physical activities, social activities, physical complaints, the quality of performance on physical activities and social activities. According to parent report, current disease activity correlated with the prevalence of physical activities, physical complaints, the quality of performance on physical activities and social activities.

Whereas, in general, within-group comparisons revealed that the HAY scales are sensitive for distinct levels of disease activity, for children with asthma and juvenile chronic arthritis more significant regression coefficients were found than for the other two groups. An explanation for this may be that in diabetes mellitus and epilepsy more disease activity does not lead directly to consequences on the prevalence of activities or problems or the quality of performance as much as it would in the other diagnostic groups. Another explanation may be that this difference is caused by the fact that current disease activity was easier to define with the available data in the groups of children with asthma and juvenile chronic arthritis. For children with diabetes mellitus a rough estimation of the number of hypoglycemias and hyperglycemias was used. For children with epilepsy, epilepsy activity and seriousness of seizures according to the parents was used. It is possible that more appropriate estimates of disease activity, such as blood sugar estimates during the day in diabetes mellitus or type of seizures combined with the number of seizures in epilepsy during the past week, could lead to more sensitive estimates of "current disease activity".

Although sensitivity for current disease activity was confirmed, especially for children with asthma and juvenile chronic arthritis, second measurements, controlling for disease activity, are necessary to assess the sensitivity for change of the HAY. In addition, objective measures, instead of parents' reports, to assess severity of symptoms are preferred in these analyses.

5.21.9 Correlation between child and parent report

As was expected, intraclass correlations between parent and child report revealed weaker correlations when the domains were less accessible to parent observation (Barbarin, 1990). This difference was however only found in healthy children. The stronger correlations between children and parents in the sample of children with a chronic illness on the quality of performance scales compared to the correlations between parents and children in the healthy sample could be explained by the possibility that parents of children with a chronic illness might keep a closer eye on their child in performing activities than parents of healthy children do. Among children with a chronic illness, parents are, in general, less positive about their child's functioning than their children, except for the assessment of general physical complaints on which parents scored less physical complaints than their children. Again, whether and when children underreport or overreport is not known. These discrepancies emphasize the need for multiple sources when conducting behavioral assessments among children.

5.21.10 Concluding remarks

The results of the main study with regard to the validity and reliability of the HAY are encouraging. In Chapter 6, results of this study will be discussed in further detail by elaborating on the operationalisation of health-related quality of life among children, testing of validity and reliability, and the use of parent and child report.

ACKNOWLEDGEMENTS

The author would like to thank the parents and children who participated so willingly in the project and also the medical specialists, GPs, and schools, for helping us to contact the parents.

CHAPTER 6: GENERAL DISCUSSION

6.1 INTRODUCTION

In this doctoral dissertation, the development of a health-related quality of life questionnaire, the HOW ARE YOU? (HAY), for children with a chronic illness, aged 7-13 years old, is described. Chronic illness has a large impact on child and family. Although the variability of outcomes is high, it is estimated that twice as many problems occur among children with a chronic illness than among healthy children. There are numerous factors that interact in complex ways to determine the adaptation of the child. The use of a theoretical framework is therefore necessary to understand the variability of consequences. Although the number of studies in this area of research is increasing, research has mainly been focused on specific areas of interest in predictors as well as in consequences. For instance, the severity of the illness is often used as a predictor. Consequences studied are often focused towards expected difficulties. In children with epilepsy, for instance, cognitive functioning has been studied relatively often, whereas in children with asthma physical functioning is a frequently studied area. The use of health-related quality of life instruments can aid in obtaining a more holistic view on the functioning of children.

Whereas the interest in health-related quality of life among children is growing, a shortage of health-related quality of life instruments has been noticed. The first part of this doctoral dissertation describes the prevalence of childhood chronic illnesses, the application of the stress and coping model developed by Maes, Leventhal & DeRidder (1996) among children with a chronic illness and the consequences of childhood chronic illness on the child's physical, social and psychological functioning. In addition, the definition and assessment of health-related quality of life among children with a chronic illness are described.

The second part of this doctoral dissertation describes the development of the HAY, including the developmental stage, the pilot study and the main study. The results of the study revealed that the HAY has a promising potential for use in research as well as in the clinical setting. In this chapter results of the study are discussed under three headings: the operationalisation of health-related quality of life among children, testing of validity and reliability, and the use of parent and child report. Finally, recommendations for future research are formulated.

6.2 SETTING UP THE HAY

Health-related quality of life assessment may assist in screening or identifying populations at risk, in assessing impact of illness or treatment, in evaluating outcomes of treatment, and it also facilitates the communication between physician, patient and parents. The following key characteristics of health-related quality of life assessment among primary school children with a chronic illness were identified as important: multidimensionality, child-friendliness, the inclusion of evaluation of importance, the incorporation of children's views as well as their parents' on similar subjects, the inclusion of positive as well as negative aspects of health-related quality of life and last but not least, the inclusion of generic as well as disease-specific aspects of life.

6.2.1 Multidimensional assessment

Particular in clinical areas, multidimensional profiles are of importance (Leger, 1988), as one health-related quality of life score may conceal the contradictory trends which can exist in the effects of illness or treatment on the functioning of children (Fitzpatrick et al., 1992; Starfield, 1987; Mulhern, Fairclough, Horowitz, Friedman, Copeland, 1989; Aaronson, 1990). When defining the multidimensionality of health-related quality of life, physical, social and psychological functioning are often included (Aaronson, 1990; Joyce, 1995; Schipper, Clinch & Powell, 1990; Patrick & Erickson, 1988; Ware 1984; Guyatt & Jaeschke, 1990). In children, these three areas were considered to be important and were included in the HAY. Interviews revealed that parents and children considered the content of the HAY as important and covering all aspects. Results of the confirmatory factor analysis confirmed that the HAY consists of the dimensions as expected and revealed acceptable correlations between the latent factors. In addition, different profiles for distinct illnesses were found.

6.2.2 A limited age range

The areas included are operationalised according to the interests and developmental level of the primary school children (Flanery, 1990; Eisen, Ware & Brook, 1979; Johnson, 1988; West, 1994). Items in the HAY are concrete and cover a relatively recent and short period of time (the previous week). In addition, pictures of faces are used to clarify the content of the "related feelings items" and to generate interest. In the pilot study, comments during the interviews revealed that children liked the pictures of sad to happy faces, although amendments had to be made in the scale range. For older children these pictures may be less appropriate as they could be seen as childish although French and French (1994) noted that also adolescents liked the "smiley faces scale" when it was used to illustrate descriptive explanations. Furthermore, for older children other aspects of daily life such as building relationships with friends, education, future plans and future career have to be included. In addition, the developmental level of adolescents permits a more

advanced understanding of concepts used such as abstract concepts like time. The developmental level of children younger than 7 years of age, on the contrary, demands other methods of self reports or even only parent reports. In self reports, pictures may have to be used in all items and other subjects increase in importance.

Considering the discussion described above, it was felt that the chosen limited age range was the only way to develop an appropriate instrument in the time available for the study. As a consequence, longitudinal research among children with a chronic illness is hindered. There is, however, no other way except to use different instruments for different age groups although these instruments should be developed according to the same principles. During childhood in particular, rapid changes in physical, mental and social abilities and interests occur. The development of the ability of children to understand, their activities during the day, their autonomy, their understanding of illness and health are rapidly changing and imply the need for different age-appropriate instruments in content and norms. New versions of the HAY are therefore needed for younger as well as older age groups.

6.2.3 Inclusion of related feelings items

The third main characteristic of health-related quality of life is the involvement of evaluation of importance. It is this evaluation which is the main distinction of health-related quality of life assessment compared to the assessment of health status or functional status. Importance was operationalised in the HAY as feelings towards limitations in functioning according to the definition of health-related quality of life of Calman (1984), who defined quality of life as the gap between achievements and expectations. In the pilot version of the HAY, this gap was assessed by asking children and parents on their happy or sad feelings towards limitations in functioning or complaints using a scale of 7 faces ranging from happy to sad. The results of the pilot study led to a reformulation which refers to a primarily cognitive process. The items were reformulated into "Does it bother you?" using a scale of 4 faces. This is in accordance with Seaberg (1990) who stated that the subjective sense of well-being can be viewed as a primarily cognitive process related to what one aspires to, feels one deserve or expects.

As the definition implies, quality of life scores should be developed consisting of a combination of the scales measuring prevalence or quality of performance with the related feelings, for example by adding or multiplying scores of both scales. Although it may be useful just to have one score, it conceals also the nature of difficulties patients experience. Individual researchers have to choose between the scoring possibilities dependent on the conclusions they want to formulate. For the study described, it was decided not to use the combined quality of life scores in the analyses. The confirmatory factor analyses could only be performed on the feelings or emotions items. Hypotheses concerning differences between groups were only

formulated for the prevalence of activities, tasks or problems and the quality of performance on activities or tasks.

We felt that children should report their own feelings. Parents were asked to report their own feelings towards their child's problems or limitations. The differences found between parents and children on an individual level can be used to estimate the differential impact of illness on parents or children (see paragraph 6.4). In this way, however, the parent version differs from the child version and in case of combination scores of prevalence or quality of performance items and related feelings items, results are not comparable.

In further research a theoretical framework should be used, so that more knowledge becomes available on how children function and how they evaluate their functioning in relation to, for example, their disease characteristics, coping abilities, internal and external resources, values and goals.

6.2.4 Inclusion of positive dimensions of life.

Health-related quality of life assessment is often oriented towards negative aspects of life. Questionnaires might therefore result in a large summation of possible complaints or limitations patients probably encounter. In the HAY also a positive feelings dimension is included, called "happiness" (Nordenfelt, 1995; Hyland, 1992). In addition, the child's prevalence of and quality of performance in physical and social activities, and cognitive tasks are inquired about in a neutral way, for instance "Have you ridden your bike during the past seven days?". The answer categories for these items include only one positive and three negative alternatives. Results from the pilot study indicated that scales including several positive categories were more difficult for children to interpret. In addition, answer categories should mainly differentiate between those children in whom we are most interested: the children with difficulties. It was therefore decided to develop items with only one positive and three negative answer categories.

6.2.5 Inclusion of generic, chronic illness and disease-specific sections.

Generic instruments can be used to assess general functioning in daily life among all children with a chronic illness to identify deviations from other diagnostic groups and from healthy children. It is important to assess the applicability of the instrument among all groups as items should reflect what they are meant to reflect (Drotar, 1994; Johnson, 1988). The modified categorical approach in studying consequences of chronic illness distinguishes generic features as well as chronic illness features and disease-specific features of illness and treatment. Generic features refer to normal daily functioning and are operationalised in the generic section of the HAY. In addition, chronic illness features are important for they are applicable to all children with a chronic illness, irrespective of their specific diagnosis. These are operationalised in the HAY in the section measuring concerns

and feelings of inferiority related to having a chronic illness. Multi-sample analysis revealed that the structure was stable in the distinct groups.

Furthermore, there are specific features of illness and treatment that ask for measures that resemble items that are discussed in clinical practice. Disease-specific sections have been developed for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. The disease-specific section for children with asthma has a good fit and good reliabilities. The epilepsy-specific section has a moderate good fit and reliabilities. The diabetes-specific section has a good fit, but low reliabilities. Reformulation and/or addition of items is recommended for this section. Furthermore, due to the small number of children with juvenile chronic arthritis included in the study, only approximate indicators of fit of the section for children with juvenile chronic arthritis can be given (Saris & Stronkhorst, 1984; Jöreskog & Sörbom, 1989; Evans, 1994). The reliabilities are good. Further research towards the sensitivity for change of these sections is needed as these sections are often needed in the evaluation of interventions.

6.3 THE TESTING OF VALIDITY AND RELIABILITY

6.3.1 Determination of construct

The first strategy to establish validity consisted of the formulation of a definition of health-related quality of life. The definition applied is derived from the definition of Calman (1984) who perceived the gap between expectations and achievements as indicative for quality of life. Literature concerning health-related quality of life of children with a chronic illness was reviewed, expert meetings were held and parents and children of the target group were interviewed. In addition, parents and children were interviewed in the pilot study on the content of the questionnaires.

6.3.2 Confirmatory factor analysis and reliability testing

The second strategy to establish construct validity consisted of confirmatory factor analysis (CFA) and reliability testing, which was used to obtain a structure of the HAY that was appropriate for each group involved. Since the related feelings items and emotional items are subjective in nature as is health-related quality of life assessment, these items were used as the basis of health-related quality of life assessment and considered to be appropriate for finding an underlying structure. The structure found was used to form related prevalence and quality of performance scales in the child version as well as in the parent version. The approach was therefore child centered with two sources of information with the focus being the child him or herself.

In the field of health-related quality of life research among children this is the first study known to have used confirmatory factor analysis, for in general, confirmatory

factor analysis is considered to be a far more appropriate method than exploratory factor analysis when the researcher has a predefined structure. Two obstacles to factor analysis have, however, been noted in quality of life research. Juniper, Guyatt, Streiner & King (1997), for example, in a study among patients with asthma, compared the inclusion of items on the basis of factor analysis with the inclusion of items on the basis of an impact method. According to the impact method items are selected that are identified most frequently and that are perceived as important by patients. For the factor analysis, highly skewed items were deleted. Both methods resulted in two partly different questionnaires. The authors concluded from this research that irrespective of their association with each other, items of functional impairment which are important to the patients should be included in a disease-specific instrument. Reliability testing and cross-sectional correlations with other measures are according to the authors important in discriminative studies, whereas for measures in a clinical trial the instrument's responsiveness and longitudinal correlations with independent measures would be key properties. The HAY can be used in discriminative as well as in evaluative studies. One of the reasons to use the related feelings items for the confirmatory factor analysis was to warrant the subjective nature of health-related quality of life.

The second problem, concerning factor analysis in quality of life research, is that factor analysis could lead to inconclusive results. One of the reasons is that cause indicators and effect indicators, which are often used together in quality of life questionnaires, are often used in the same analysis (Fayers & Hand, 1997). For example, tiredness in one group can cluster with illness or treatment characteristics, whereas in another group tiredness can cluster with depression. The use of causal indicators in factor analysis is therefore not recommended (Fayers & Hand, 1997). This was a second reason to apply the confirmatory factor analysis on the related feelings and emotional items.

A critical comment on our study might be that the related feelings items were answered irrespective of whether the child had experienced problems. The factor structure is therefore based on feelings that are related to the problem experienced, as well as on related feelings towards imagined problems. This method, however, was the only way to use the related feelings items for the factor analyses as a large number of missing data would otherwise occur. A second argument is that the current method leads to a structure based on the opinions of all children, irrespective their illness or their limitations. Multi-sample analyses led to the conclusion that the structure was stable in all groups. As a result, it is acceptable to compare groups on these dimensions.

Another critical comment could be that the parents' version of the HAY was based on the item structure found among children. It was felt that this was the right way to develop a health-related quality of life instrument for measuring the child's

quality of life as the goal of parent report was to assess parents' views on the same issues as those assessed in the child version. Reliability estimates confirm the internal consistency of the dimensions in the parent version.

Confirmatory factor analysis and multi-sample analyses revealed, in general, a good fit for the structure of the generic as well as the chronic illness sections and the disease-specific sections of the HAY. Although it was suggested that it is important to assess the social domain in children with a chronic illness, the assessment of social functioning in clinical studies is relatively scarce. Results of the confirmatory factor analysis in the main study revealed that the social problems dimension had to be deleted. Social functioning is therefore only assessed by the prevalence of and quality of performance in social activities and the child's or parent's related feelings towards limitations in this area. Although results of the pilot revealed no differences between groups on the social problems dimension, further research is needed on the necessity of this dimension in addition to the dimensions measuring social activities.

6.3.3 Concurrence with the CBCL and the CATIS

As a third strategy to establish construct validity, cross-structure analysis was performed, including the testing of hypotheses concerning the relations with other instruments and hypotheses concerning differences between groups. Concurrent validity was tested by assessing the correlations between dimensions of the HAY with the Child Attitude Toward Illness Scale (CATIS) (Austin & Huberty, 1993) and the Child Behavior Checklist (CBCL) (Verhulst, Koot, Akkerhuis & Veerman, 1990; Verhulst, Ende & Koot, 1996). When testing concurrent validity, it is preferable to choose a well-validated instrument that is hypothesized to assess identical constructs. At the time the research started, there were however no gold standards available.

It was therefore decided to use the Child Attitude Toward Illness Scale (CATIS), which was found to have good validity and reliability in children with asthma and epilepsy and was easy to administer (Austin & Huberty, 1993), to examine the validity of the two dimensions of the HAY measuring the child's concerns and feelings of inferiority towards illness. First, the CATIS was translated for use in the Netherlands. Answer categories were changed so that each item had the same answer categories from never to always. Items were therefore reformulated. Results of the confirmatory factor analysis and the Cronbach's alphas revealed a valid and reliable instrument for use in the Netherlands. As it was the first time that the CATIS had been used in the Netherlands, more research does need to be done on the validity and reliability of the CATIS, for example by testing the stability of the instrument among groups and hypotheses concerning differences between groups.

The results of the main study indicated moderate correlations between the disease-specific section of the HAY and the CATIS. In validating health-related quality of life measures when there is no criterion standard, correlations with related measures greater than 0.5 are seldom observed and should not be expected (Hillers et al., 1994). Correlations ranging from .52 to .59 were observed between the dimensions measuring concerns and feelings of inferiority towards illness of the child version of the HAY and the CATIS, which suggests that the HAY succeeds in capturing feelings of inferiority and concerns related to having a specific chronic illness.

The CBCL was used to estimate the concurrence between the quality of performance scales and physical complaints scales of the HAY with comparable constructs measured by the CBCL. The CBCL is based on two large standardized samples, one consisting of children referred for mental health treatment and the other of children who had not been referred. Good validity and reliabilities for the CBCL were found when tested in a large Dutch sample and the CBCL is often used in studies among children with a chronic illness (Verhulst, Koot, Akkerhuis & Veerman, 1990; Verhulst, Ende & Koot, 1996; Perrin, Stein & Drotar, 1991).

Despite these advantages, limitations of the CBCL have also been reported such as a possible bias in interpreting data concerning physical symptoms, a limited sensitivity when identifying mild adjustment problems of the sort most often encountered in children with chronic physical illness, and an incomplete and potentially misleading assessment of social competence as not only competence but also the involvement in social activities is included (Perrin, Stein & Drotar, 1991; Harris, Canning & Kelleher, 1996). As there were no other instruments available for children measuring physical, social and cognitive functioning with such a wide application, good validity and large norm groups to assess psychosocial problems, it was decided to include the CBCL in the study. Further research with the use of the CBCL among children with a chronic illness and obtaining norm groups among these children is however needed.

Only moderate correlations could be expected between the quality of performance scales of the HAY and the competence scales of the CBCL. Correlation testing between the quality of performance scales and the physical complaints scale of the HAY with the competence scales and the physical symptoms of the CBCL revealed moderate correlations. The highest correlations were found between the scales measuring cognitive competence, and physical complaints. A possible explanation for this might be the fact that the physical and social competence scales of the CBCL measure not only quality of performance but also the involvement of children in physical and social activities. These scales are thus less comparable than the scales measuring cognitive competence and physical complaints. The correlations revealed concurrence between the scales, which supports the validity of the HAY.

New instruments measuring health-related quality of life that have been tested on validity and reliability among children have become available during the past five years in which also the HAY was developed. Concurrent validity of the HAY should therefore be tested using generic instruments, such as the TNO-AZL-Child Quality of Life questionnaire (TACQOL) (Verrips et al., 1997; Vogels et al., 1998) or, for the disease-specific sections, disease-specific instruments such as the Child Asthma Questionnaire (CAQ) (French & Christie, 1994) or the Paediatric Asthma Quality of Life Questionnaire (PAQLQ) (Juniper et al., 1996a). It must however be noted that almost all instruments are relatively new and have their own merits, which are different from the HAY.

6.3.4 Discriminative properties

Discriminative properties of the HAY were examined by assessing differences between children with a chronic illness and healthy children as well as between groups of children with asthma, diabetes mellitus, epilepsy, juvenile chronic arthritis and healthy children. To prevent the results being found by chance because of multiple testing, a probability level of .01 or .001 was used. As differences are also bound to gender, gender was used as a factor. Age was used as a covariate.

Part of the hypotheses with regard to differences between children with a chronic illness and healthy children were confirmed. Significant differences in the expected direction in the child version were found for the prevalence of social activities and the quality of performance on physical activities. In addition, children with a chronic illness scored more negative feelings on limitations in social activities than healthy children did.

Significant differences in the expected direction in the parent version were found for the prevalence of social activities, general physical complaints and the quality of performance on physical activities, cognitive tasks and social activities. Like their children, parents of children with a chronic illness had more negative feelings about limitations in social activities than parents of healthy children had. Rather than the prevalence of activities or tasks, according to their parents, the quality of performance seems to be affected in children with a chronic illness.

Part of the hypotheses with regard to differences between each of the disease-specific groups, e.g. children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis, and the healthy group were confirmed using either child report or parent report.

On physical activities, more limitations were found among children with asthma and juvenile chronic arthritis, as was hypothesized. Children with diabetes mellitus even had a higher prevalence of physical activities than healthy children, possibly as a consequence of the fact that exercise and a balance between exercise, diet and rest,

is recommended for children with diabetes mellitus. This finding should, however, be examined in further studies. Children with diabetes mellitus and children with asthma experienced more physical complaints than healthy children.

On cognitive functioning, children with epilepsy scored a lower quality of performance as was hypothesized, according to parent report. None of the groups scored, however, lower on the prevalence of cognitive tasks although school absenteeism is reported to be higher among children with a chronic illness. The scale measuring the prevalence of cognitive tasks is probably not sensitive enough to measure school absenteeism just for part of the week. In addition, children who stay home because they are ill, might do their schoolwork at home and thus perform their cognitive tasks.

On social activities, children with asthma scored significantly more limitations than healthy children, as was hypothesized. The fact that children with epilepsy did not score lower on these activities is not easy to explain. A possible explanation might be that it is not the social activities but rather the social relationships which are more problematic in children with epilepsy (Hoare & Kerley, 1991). As the social activities scales also measure the involvement in physical activities, one would also expect children with juvenile chronic arthritis scoring lower than healthy children which wasn't found in this study. It is possible that children with juvenile chronic arthritis have more visible limitations in physical activities than children with asthma, so that it is more easy to explain and join in other social activities or in another role. Further research towards the social activities and social relationships among children with a chronic illness is warranted.

No hypotheses were formulated concerning feelings of happiness or feelings towards complaints or limitations as these are individual matters, depending on many variables as described in Chapter 1, such as coping, or social support, goals or values a child may hold. As was stated above, the assessment of functioning as well as related feelings or happiness on an individual level is recommended. When children experience limitations or complaints and consider these as bothersome, these issues should be the main targets of treatment, either in relieving symptoms or restructuring expectations. A significant finding is, however, that on a group level, children with juvenile chronic arthritis as well as their parents scored more negative feelings towards limitations in physical activities and social activities than healthy children or their parents. This finding reveals that these areas are of concern to children with juvenile chronic arthritis and their parents so that attention should be directed to these areas in clinical practice as well as in research.

6.3.5 Evaluative properties

Like the discriminative properties, the evaluative properties of a health-related quality of life questionnaire are also important. Evaluative properties are of major

importance when one wants to use the HAY in clinical trials, psychological interventions or evaluation in clinical practice. Responsiveness to change, e.g. how well scores distinguish between improved and unimproved patients (Deyo, Diehr & Patrick, 1991) is therefore an important criterion. A study among 77 children with asthma indicated that the scores on most prevalence and quality of performance scales of the HAY child version as well as the parent version changed when the children's actual asthma status changed (le Coq, Boeke, Bezemer, Colland, Bruil & van Eijk, 1998a; le Coq, Boeke, Bezemer, Colland, Bruil & van Eijk, 1998b). In addition, the HAY has been used among children with diabetes mellitus in an intervention study and is currently used among children with juvenile chronic arthritis to evaluate interventions, but these results have not yet been analyzed. As results from a second measurement of the HAY for the total group of children with a chronic illness are not available at present, it was decided to analyze the relation between current disease activity as reported by the parents and levels of functioning.

Responsiveness to severity was assessed by correlating current disease activity with the level of child's functioning after controlling for age and gender. An important obstacle for measures of responsiveness is the accuracy of the measure of "severity". Severity can only be assessed within distinct groups, as underlying physical symptoms, such as hypoglycemias or breathlessness, are often very distinct from each other. In this study severity was assessed by asking parents to indicate disease-specific symptoms. As such, the term, current disease activity, was considered to be more appropriate than severity. Results indicated a better sensitivity of the HAY to current disease activity in children with asthma and juvenile chronic arthritis, but these results may be biased by a better definition of current disease activity within these two groups.

Studies often rely on parent-reported severity, but a bias in favor of greater agreement on parent-reported health-related quality of life and severity may therefore occur (Fritz & Overholser, 1989). To improve understanding of the nature of the relation between physiological data and health-related quality of life data, more objective physical data on the nature of the child's illness are needed when studying the relationship between severity and parent-reported or self-reported level of functioning.

6.4 THE USE OF CHILD AS WELL AS PARENT REPORT

Many studies revealed that reports of patients and proxies or parents and children do not correlate highly on health-related quality of life issues. In particular on issues that are difficult to observe, differences occur. Various hypotheses concerning the disagreement of children and parents have been proposed in a review of the literature by Stone & Lemanek (1990): (1) children's perceptions are accurate but

they do not report certain behaviors, subjective symptoms, to others; (2) perceptions are different but they reflect actual behavioral differences in specific settings; (3) dissimilar reports are a function of symptom type and disorder; 4) parents' reports are influenced by a negative halo effect whereby parents tend to endorse many negative characteristics of their children.

There is much debate about whether to use parent or child report in assessing health-related quality of life. It has, however, been stated before that one of the most essential characteristics of health-related quality of life is the individual's evaluation of the situation. Parents' reports on these issues would be difficult to interpret, as children's feelings are difficult to observe for parents and parents' report would reflect the parents' interpretation of the child's feelings parents mixed up with their own feelings rather than the child's feelings. We therefore decided not to use parent report on the feelings of their child but to assess parents' own feelings. An exception is the scale measuring the prevalence of happy feelings, in which parents are asked whether their child had happy feelings. These feelings are probably more easy to observe, but it would be more in line with the previous statements to ask parents for their own happy feelings.

The ICC on the prevalence and quality of performance scales for children with a chronic illness ranged from .36 to .65, for healthy children the ICC ranged from .27 to .59. A meta-analysis on consistency between informants of behavioral and emotional problems revealed that ratings were more consistent for 6- to 11-year-old children than for adolescents (Achenbach, McConaughy & Howell, 1987). Other consistency rates may therefore be revealed when developing a health-related quality of life instrument for older children (see also Johnson, Silverstein, Rosenbloom, Carter & Cunningham (1986).

Studies suggest that both reports have their own value (Rosenbaum, Cadman & Kirpalani, 1990; Eisen, Ware & Brook, 1979; Achenbach, McConaughy & Howell, 1987; La Greca, 1990a). We stated earlier that only the child itself can inform us about the limitations or complaints he or she is experiencing and what feelings occur with these limitations or complaints (Maes & Bruil, 1994). Parents may report their view about heir child's functioning and how they feel about limitations or complaints. Disagreements between parents and children should be used complementarily as they can highlight variations in judgements made by the child and the parent about the child's functioning in various situations. Such differentiation in turn can provide more differentiated foci for interventions and outcome evaluations (Achenbach, McConaughy & Howell, 1987). In studies involving children, parents may serve as a primary data source when necessary, for instance when children are too young, mentally disabled or too sick. It is clear that data derived in this indirect manner should be interpreted with great caution (see also Aaronson, 1990).

6.5 RECOMMENDATIONS FOR FUTURE RESEARCH

Results of the study and the subsequent discussion revealed the following suggestions for future research on the HAY and, more broadly, health-related quality of life among children with a chronic illness:

1. The validity and reliability of the disease-specific subsection of the HAY for children with diabetes mellitus has to be adapted and retested in a group of children with diabetes mellitus and their parents.
2. The validity and reliability of the disease-specific section for children with juvenile chronic arthritis has to be confirmed in a larger sample of children with juvenile chronic arthritis and their parents.
3. Disease-specific sections should be developed for children with chronic illnesses other than asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. During the development of the HAY, disease-specific sections for children with cancer and chronic renal failure have been developed but have not been tested in a validation study yet.
4. The sensitivity to change needs to be investigated further whereby preferably an objective parameter of disease activity is also included to permit the HAY to be used in clinical trials.
5. Versions for children younger than 7 years of age and older than 14 years old that fit their interest and suit their developmental level have to be developed.
6. In the Netherlands, many children have a non-Dutch ethnic background. The children included in the study had, for the most part, a Dutch ethnic background. To be able to help children from other cultural or ethnic backgrounds as well as their parents, the use of the instrument should be tested in these groups too.
7. International research to validate the use of the HAY in other countries is important to be able to do international research and compare results from different countries. Although it is estimated that 20% of children is chronically ill, a large number of diseases are rare. We recommend carrying out more international research among these children with instruments that are valid and reliable in other cultures and countries.
8. Further research is needed on the validation of the CATIS in the Netherlands, including testing hypotheses concerning differences between groups and comparisons with other instruments. In addition, the use of the CBCL among children with a chronic illness needs to be explored further.
9. More research should be directed towards predictive variables of health-related quality of life. In addition, health-related quality of life among children should be studied using a theoretical framework in which the modified categorical approach is used and a holistic view of children is obtained. Only then does more information become available about how to identify children at risk and where precisely more help is needed to prevent negative consequences.

Health-related quality of life assessment in pediatric psychology is in its infancy, but opportunities to obtain more knowledge about, and understanding of health-related quality of life among children with a chronic illness are increasing. The HAY can be used to assess health-related quality of life among children with a chronic illness aged 7 to 13 years old. The measure includes a generic as well as a chronic illness section and disease-specific sections for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. The HAY provides a profile of the functioning of the child covering physical, social and psychological consequences of illness, measuring the child's limitations or complaints as well as its related feelings. Negative as well as positive aspects of health-related quality of life are included. Parents' views as well as those of their child are obtained on similar subjects so that the measure captures both views. Personal feelings or evaluations are however only obtainable from self report. The measure is child-friendly in the language, the concepts and the layout used. Recommendations as described above should lead to a better use of health-related quality of life assessment and further development of health-related quality of life assessment in pediatric clinical practice and research.

SUMMARY

Chronic illness in primary school children

Chronic illnesses remain a challenge to medical science for these illnesses are either debilitating, fatal or can be managed successfully although requiring life-long treatment. Childhood chronic illness (Chapter 1) is defined as “a physical condition that affects children for extended periods of time, often for life. The illnesses can be ‘managed’ to the extent that a degree of pain control or reduction in attacks (of asthma), bleeding episodes (in hemophilia) or seizures can generally be achieved. However they cannot be cured” (Eiser, 1990b; p.3).

Despite the fact that medical care has been improved, prevalence estimates of childhood chronic illness are stable or even increasing. It has been estimated that one-fifth of all children are chronically ill and that half of this group has moderate or serious limitations in daily life. Psychosocial problems seem to occur twice as often in the group of children with a chronic illness than in the group of healthy children, although the variability of consequences described is large. The stress and coping model developed by Maes, Leventhal and DeRidder (1996) was used to describe important variables in assessing the risk or resistance factors.

Variables, studied often, are illness characteristics, which refer to generic as well as disease-specific features of illness. In this doctoral dissertation the *modified categorical approach* is used. In this approach, general illness characteristics as well as unique features are incorporated. Four illnesses have been selected to study the consequences of childhood chronic illness, these being asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. These illnesses are similar in several dimensions resulting in common experiences that may influence the child’s functioning. The illnesses are viewed as nonfatal, children with these illnesses are generally able to attend normal schools and participate in social activities. Furthermore, their management requires daily treatment, diet or exercise. In addition, each illness also has unique aspects in paroxysmality, brain involvement and physical impairment that may influence a child’s daily experiences and their functioning in normal daily life.

Variables influencing the consequences of chronic illness include further important life events, illness-related events, demographic characteristics such as age and gender, goals or values of the children, external and internal resources and coping. The consequences of illness refer to the physical functioning, including physical complaints, physical limitations and daily treatment tasks, social functioning and psychological functioning, including cognitive as well as emotional and behavioral outcomes.

Consequences of childhood chronic illness

In Chapter 2, a review of clinical studies among children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis on these dimensions of functioning is given. Based on these studies, it was concluded that children with asthma experienced more physical complaints and more limitations in physical activities as well as in social activities and had more school absenteeism compared to healthy children. Children with diabetes mellitus were reported to have more problems in social functioning than healthy children. Children with epilepsy appeared to experience more limitations on cognitive tasks and social functioning than healthy children. Children with juvenile chronic arthritis were reported to experience more physical complaints and limitations in physical activities and social functioning and more school absenteeism than healthy children.

Reviewing the dimensions studied within each group, it could be concluded that expected difficulties in specific illnesses were more extensively studied than other dimensions. A holistic view on the functioning of the child is therefore missing and unexpected problems may thus easily be overlooked. In addition, in most studies, problems experienced by children are assessed in comparison with a healthy group or a normative sample. Most studies used therefore generic instruments whereas the use of illness-specific scales was rare and only used in separate studies. Although parent report was often guided by child report, these reports were seldom used complementarily. In just a few cases a physician's report or teacher's report was added.

All these studies point to the differing areas of daily life upon which a chronic illness may have a large impact. It was concluded that "health-related quality of life" assessment is necessary to obtain a holistic view of the functioning of children with a chronic illness and that parent as well as child reports are needed.

Health-related quality of life among children with a chronic illness, the definition and assessment

Chapter 3 describes the definition and assessment of health-related quality of life in children with a chronic illness. Health-related quality of life is considered to refer to the functioning of children in the physical, social and psychological dimensions of life, and the gap between their achievements and expectations related to their functioning. The functioning of children is described in terms of prevalence of activities or tasks, symptoms, problems or emotions and the quality of performance in activities. The gap between achievements and expectations is defined by the appraisal of difficulties related to the physical, social and psychological functioning.

Reviewing important characteristics of health-related quality of life assessment among children with a chronic illness revealed the following criteria for a health-related

quality of life instrument: (a) the measure should be multidimensional covering the physical, social and psychological functioning and report a profile of the functioning of the child; (b) the instrument has to measure the functioning of the child as well as related feelings towards limitations or problems in functioning; (c) the measure should assess positive as well as negative emotions; (d) the instrument should consist of a generic as well as a disease-specific section; (e) the reference group should include healthy children as well as children with a chronic illness, in relation to the target group of this doctoral dissertation, including children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis; (f) the measure should include the parents' views as well as their child's on similar subjects so that the measures captures both views. Feelings related to problems and emotions are however only obtainable from self-report; (g) the instrument should be child-friendly in how it is written and in the concepts used as well as in the layout.

Due to the scarcity of health-related quality of life instruments and the relative recency of most instruments, the experience with health-related quality of life assessment among children is very limited. A review of current health-related quality of life instruments revealed that all current instruments have merits that have to be checked when looking for a specific measure and all have some apparent relationship with the definition of health-related quality of life. It appeared however that none of the existing instruments fulfilled all the criteria formulated. It was therefore decided to develop the HOW ARE YOU? (HAY) a health-related quality of life questionnaire for children, aged 7-13 years old.

The development of the HOW ARE YOU? (HAY), results of the pilot study

Chapter 4 describes the developmental stage of the HAY and the results of a pilot study on the validity and reliability of the HAY. The content of the HAY was based on literature, interdisciplinary discussions, interviews with parents and children and then reviewed by a panel of experts. The HAY consists of a generic section, applicable to all children, a chronic illness section applicable to all children with a chronic illness and four disease-specific sections developed for children with respectively asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis.

Preliminary data on utility, validity and reliability were obtained in the pilot study in which 89 children with a chronic illness and 134 healthy children and their parents were involved. In addition, the concurrence between parents and children was examined. Data on utility were obtained by interviewing parents and children about the relevance of items and amendments that should be made. Validity was tested by means of confirmatory factor analyses and the testing of hypotheses concerning the relation between the Child Behavior Checklist (Verhulst, Koot, Akkerhuis & Veerman, 1990) and the HAY and differences between groups. Cronbach's alphas were used to estimate reliability.

Results revealed that parents as well as children could complete the HAY in a reasonable period of time. The average time children needed to complete the HAY needed was 30 minutes. Amendments of items and answer categories were, however, necessary.

An acceptable fit for the generic section and good reliabilities for the separate generic dimensions was found. The chronic illness section for all children with a chronic illness, measuring emotional response to having an illness, could however not be evaluated because the dichotomous items were highly skewed. The disease-specific sections for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis measuring disease-specific physical complaints and daily management tasks, had moderate to high reliabilities, but could not be assessed on structure because of small sample sizes. For children with asthma and epilepsy a daily management tasks dimension was missing and had to be added.

Testing the hypotheses with regard to correlations with the CBCL revealed moderate correlations, ranging from .30 to .51, indicating concurrence between both instruments neither of which were expected to be high as no gold standards were used.

Comparisons between groups revealed that most hypotheses in respect of the differentiation between groups were confirmed. Moderate correlations between child and parent report revealed that both versions should be used complementarily.

Although the results of the pilot study were encouraging, it appeared that more research was needed. Revisions of items and answer categories as well as addition of dimensions were necessary. In addition, the validity and reliability of the HAY had to be tested on a larger sample of children. This was, in particular, of importance for estimating the validity of the chronic illness and disease-specific sections.

The validity and reliability of the HAY

In Chapter 5 the results of the main study are described. In this study the validity and reliability of the revised version of the HAY were assessed involving a large group of children with a chronic illness and their parents (N=577) and healthy children and their parents (N=418). Results revealed that the HAY could be answered alone by most children.

Construct validity was examined by analysis of the meaning and operationalisation of the construct, confirmatory factor analysis, concurrence with other instruments and testing hypotheses concerning differences between groups. Confirmatory factor analyses and reliability testing were performed on the related feelings items and the emotional items. The structure found among these items was used to form the

related prevalence and quality of performance scales of the child version as well as the parent version.

Confirmatory factor analyses and reliability testing revealed that the generic section of the HAY consists of five dimensions with a good fit and good reliabilities, measuring physical activities, cognitive tasks and social activities, physical complaints and happiness. The Cronbach's alphas in the child version ranged from .77 to .86. The Cronbach's alphas in the parent version ranged from .86 to .93. The chronic illness section is applicable to all children with a chronic illness and includes two dimensions with a good fit and good reliabilities, measuring concerns and feelings of inferiority related to having a chronic illness. The Cronbach's alphas of these dimensions were respectively .80 and .74 in the child version and .86 and .66 in the parent version. The disease-specific sections are different for each group. The asthma-specific section consists of two dimensions with a good fit and good reliabilities called "asthma-specific physical complaints" and "daily management". The Cronbach's alphas of these dimensions were respectively .77 and .76 in the child version and .83 and .78 in the parent version. The diabetes-specific section consists of three dimensions, including "physical complaints related to a hyper", "physical complaints related to a hypo" and "daily management". The Cronbach's alphas of these dimensions were respectively .58, .63 and .63 in the child version and .62, .67 and .71 in the parent version. Although the model had a good fit, reliabilities of the dimensions were considered to be unsatisfactory. The epilepsy-specific section consists of three dimensions had a good fit and good reliabilities, including "epilepsy-related physical complaints", "seizures" and "daily management". The Cronbach's alphas of these dimensions were respectively .74, .78 and .70 in the child version and .72, .83 and .76 in the parent version. The arthritis-specific section also consists of three dimensions with good reliabilities and a reasonable fit, including "arthritis-related complaints", "limitations" and "daily management". The Cronbach's alphas of these dimensions were in the child version were respectively .73, .74, .76 and .81, .73 and .74 in the parent version. The results of the confirmatory factor analysis were, however, not stable in this group because of small sample size.

Hypotheses concerning the correlations with other instruments were tested revealing that the correlations between the child version of the HAY scales measuring "concerns" and "feelings of inferiority" and a revised version of the Child Attitude Toward Illness Scale (CATIS) (Austin & Huberty, 1993) were significant, being .53 and .59, but also revealed unique components of both instruments.

Correlation between the scales measuring physical, cognitive, social competence and physical complaints of the Child Behavior Checklist (CBCL) (Verhulst, Koot, Akkerhuis & Veerman, 1990; Verhulst, Ende & Koot, 1996) and the quality of performance scale and the physical complaints scale of the parent version of the HAY revealed that correlations on cognitive competence and physical complaints

were the highest. The correlations on the physical and social competence scales were low. This discrepancy may be explained by the fact that the CBCL physical and social competence scales also measure involvement in activities, whereas the HAY quality of performance scales only measure competence. The concurrence between the CBCL and HAY supports the validity of the HAY as no high correlations could be expected when gold standards do not exist.

Significant differences in the expected direction were found for the prevalence of social activities and the quality of performance on physical activities according to child report. Children with a chronic illness scored significantly lower than healthy children on the prevalence of social activities and the quality of performance on physical activities. In addition, children with a chronic illness scored more negative feelings on limitations in social activities than healthy children did. Contrary to our expectations, children with a chronic illness did not report more general physical complaints compared to healthy children. One explanation for this finding might be that children with a chronic illness are more used to general physical complaints and thus ignore them more often. This finding stresses the importance of the assessment of disease-specific complaints because scales measuring general complaints will probably not detect fully the relevant physical symptoms that such children experience in daily life.

Significant differences in the expected direction in the parent version were found for the prevalence of social activities, general physical complaints and the quality of performance on physical activities, cognitive tasks and social activities. Like their children, parents of children with a chronic illness had more negative feelings about limitations in social activities than parents of healthy children had. No significant results were, however, found on the prevalence of physical activities and cognitive tasks. These results might be explained by the fact that, in general, rather than the prevalence of physical activities, the quality of performance in physical activities is affected for children with a chronic illness like the illnesses included in the study.

In relation to differences between disease-specific groups, the following results were found on the distinct dimensions. According to child as well as parent report, more limitations on physical activities were reported by children with asthma and juvenile chronic arthritis than by healthy children, as was hypothesized. In contrast, children with diabetes mellitus had a higher prevalence of physical activities than healthy children, according to both child report and parent report. Children with diabetes mellitus and children with asthma experienced more physical complaints than healthy children according to parent report. The illness characteristics of diabetes mellitus might explain the higher prevalence of complaints such as headaches or feeling nauseous, as these complaints can accompany hypoglycemia or hyperglycemia. In contrast to what was expected, children with juvenile chronic arthritis did not experience more general physical complaints. A possible

explanation might be that most studies on which the hypothesis was based included disease-specific physical complaints and it is in this area that problems are apparent.

On cognitive functioning, children with epilepsy scored a lower quality of performance according to parent report, as was hypothesized. None of the groups scored, however, lower on the prevalence of cognitive tasks although it was hypothesized that school absenteeism, and therefore also the prevalence of cognitive tasks, would be higher among children with a chronic illness. The scale measuring the prevalence of cognitive tasks is probably not sensitive enough to measure school absenteeism just for part of the week. In addition, children who stay home because they are ill, might do their schoolwork at home and thus perform their cognitive tasks.

On social activities, children with asthma scored significantly more limitations than healthy children, as was hypothesized. The fact that children with epilepsy and children with juvenile chronic arthritis did not score lower on these activities is not easy to explain.

No hypotheses were formulated about feelings of happiness or feelings towards complaints or limitations as these depend on many variables such as goals or values a child may hold. No significant differences were found for the prevalence of happy feelings of the children between the groups. Differences between disease-specific groups and healthy children were found on feelings towards limitations in physical activities, on which children with juvenile chronic arthritis and their parents scored lower than healthy children and their parents. In addition, children with asthma themselves as well as children with juvenile chronic arthritis and their parents scored more negative feelings towards limitations in social activities than healthy children or their parents.

As was stated above, the assessment of functioning as well as related feelings or happiness on an individual level is recommended. When children experience limitations or complaints and estimate these as bothersome, these issues should be the main targets of treatment, either in relieving symptoms or restructuring expectations.

As far as differences between boys and girls are concerned, girls generally reported a lower prevalence of physical activities, a higher prevalence of physical complaints and more negative feelings about physical complaints than boys which is in accordance with previous studies. There were no significant interaction effects between illness and gender.

The ability of the HAY to differentiate within groups on the level of disease activity was assessed by relating current disease activity as reported by the parents to the level of functioning of the child after controlling for age and gender. Results

revealed that, in particular for children with asthma and children with juvenile chronic arthritis, current disease activity concurs with limitations in functioning or complaints as reported by parents and children. An explanation might be that current disease activity was easier to define with the available data in the groups of children with asthma and juvenile chronic arthritis. Second measurements, controlling for disease activity, are necessary to assess the sensitivity for change of the HAY.

Intraclass correlations between child and parent report revealed concurrence between these two reports, which was higher for the prevalence than for the quality of performance scales in healthy children. This difference may be explained by the fact that assessment of quality of performance involves a more individual, subjective interpretation. In addition, the intraclass correlations were higher for the group of children with a chronic illness on quality of performance scales than for the healthy children. A possible explanation is that children with a chronic illness may be more supervised by their parents than healthy children. In particular among children with a chronic illness, parents scored more limitations in physical, cognitive and social activities and less happy feelings than their children, but on the contrary also fewer physical complaints than their children. These results may indicate that parents have, in general, a more pessimistic view about their child's functioning. In addition, the results may also indicate that internal processes, as physical complaints are more difficult to detect for parents. We recommend that the reports should be used complementarily.

Conclusion

In conclusion, results of the study revealed that the HAY is a valid and reliable instrument for the assessment of health-related quality of life among children with a chronic illness aged 7 to 13 years old for research purposes. In clinical practice it can be used to guide the communication between physician and child and parents. The measure includes a generic as well as a chronic illness section and disease-specific sections for children with asthma, diabetes mellitus, epilepsy and juvenile chronic arthritis. The HAY provides a profile of the functioning of the child covering physical, social and psychological consequences of illness, measuring the child's limitations or complaints as well as its related feelings. Negative as well as positive aspects of health-related quality of life are included. Parents' views as well as those of their child are obtained on similar subjects so that the measure captures both views. Personal feelings or evaluations are however only obtainable from self-report. The measure is child-friendly in the language, the concepts and the layout used.

SAMENVATTING

Chronische ziekte bij kinderen

In dit proefschrift wordt de ontwikkeling van een kwaliteit van leven vragenlijst, de HOE GAAT HET? (HOW ARE YOU?, HAY), beschreven. Deze vragenlijst is ontwikkeld voor kinderen van 7 tot en met 13 jaar met een chronische ziekte. Chronische ziekten bij kinderen zijn in dit proefschrift gedefinieerd als "ziekten die kinderen langdurig treffen, veelal voor hun hele leven. De ziekten kunnen worden behandeld in die zin dat een bepaalde mate van pijn of vermindering van aanvallen (astma en epilepsie), bloedingen (bij hemofilie) kan worden bereikt. De ziekte kan echter niet genezen" (zie Eiser, 1990b) (Hoofdstuk 1).

Ondanks een betere medische zorg en een toegenomen kennis van ziekten, is de prevalentie van chronische ziekten gestegen. Uit epidemiologische studies blijkt dat ongeveer een vijfde van de kinderen chronisch ziek is en dat de helft van deze groep ernstige beperkingen in het dagelijks leven kent. Tevens blijkt uit onderzoek dat psychosociale problemen in deze groep tweemaal zoveel voorkomen dan in een groep gezonde kinderen, hoewel er een grote variabiliteit in de waargenomen problemen is. Het stress- en copingmodel van Maes, Leventhal en De Ridder (1996) wordt in dit proefschrift gebruikt om de variabelen die van belang zijn bij het ontstaan van problemen te beschrijven.

Ziektekenmerken, zoals progressiviteit van de ziekte, aangevalsgewijs optredende klachten en zichtbaarheid voor anderen, zijn veelvuldig onderzochte variabelen in verband met het ontstaan van problemen. In dit proefschrift worden zowel algemene als ziektespecifieke kenmerken, kenmerken die betrekking hebben op een bepaald ziektebeeld, besproken. Aan het onderzoek hebben kinderen met astma, diabetes mellitus, epilepsie en juveniele chronische artritis (in het vervolg reuma genoemd) deelgenomen. Voor al deze ziekten geldt dat zij niet als dodelijk worden gezien, kinderen kunnen in het algemeen naar gewoon basisonderwijs en deelnemen aan sociale activiteiten. De ziekten vereisen dagelijkse zelfzorg, bijvoorbeeld door het gebruik van medicatie, insuline, het moeten doen van oefeningen en het zich houden aan bepaalde gedragsregels. Daarentegen zijn er ook verschillen, bijvoorbeeld in het aanvalsgewijs optreden van de symptomen en de soort fysieke symptomen, zoals benauwdheid bij astma, hypo's of hyper's bij kinderen met diabetes mellitus, aanvallen bij epilepsie, of pijn bij reuma.

Het is inmiddels achterhaald om uitsluitend ziektekenmerken als bepalend te zien voor het ontstaan, of juist niet ontstaan, van negatieve fysieke en psychosociale gevolgen. Andere variabelen die eveneens bestudeerd zouden moeten worden zijn onder meer andere levensgebeurtenissen, ziektegerelateerde gebeurtenissen,

demografische kenmerken, zoals leeftijd en sekse, doelen of waarden van het kind, persoonlijkheid, de manier van omgaan met de ziekte en sociale steun.

De consequenties van de ziekte kunnen worden onderverdeeld in drie domeinen van functioneren, namelijk fysiek functioneren, sociaal functioneren en psychologisch functioneren. Onder het fysiek functioneren worden fysieke klachten, fysieke beperkingen en zelfzorg verstaan. Onder sociaal functioneren worden sociale activiteiten en sociale problemen verstaan. Psychologisch functioneren omvat zowel cognitief functioneren als emotionele problemen. Gezondheidsgerelateerde kwaliteit van leven omvat al deze gebieden van functioneren.

Het functioneren van kinderen met een chronische ziekte

In hoofdstuk 2 wordt een overzicht gepresenteerd van de conclusies van studies verricht bij kinderen met astma, diabetes mellitus, epilepsie en reuma. De studies werden geselecteerd uit de databestanden van Psyclit en Medline CD-Rom in de periode 1987 tot 1997 en waren gericht op het fysiek, cognitief, sociaal en emotioneel functioneren van kinderen.

Op basis van dit overzicht werden hypotheses opgesteld aan de hand waarvan kan worden nagegaan of the HAY voldoende differentieert tussen de vier groepen (zie hiervoor hoofdstuk 4 en 5).

Uit dit overzicht blijkt dat de uitkomsten van studies erg variëren. Dit is onder meer te wijten aan de verschillende methoden van onderzoek, de subgroepen die bestudeerd worden, de variabelen die in het onderzoek worden meegenomen, het instrumentarium en de gekozen vergelijkingsgroepen of normgroepen. Daarnaast blijkt dat studies veelal gericht zijn op die gebieden van functioneren waarin problemen verwacht worden, bijvoorbeeld fysiek functioneren bij kinderen met astma of cognitief functioneren bij kinderen met epilepsie. Hierdoor ontbreekt een totaalbeeld van functioneren op meerdere gebieden. Tenslotte blijkt dat slechts in weinig studies zowel ouders als kinderen naar hun mening over het functioneren van het kind gevraagd werd. Dit is mogelijk ook te wijten aan een gebrek aan instrumentarium dat zowel door ouders als kinderen gebruikt kan worden. Bovendien werden dikwijls algemene instrumenten gebruikt, die ontwikkeld zijn voor gezonde kinderen. Bij het gebruik van algemene vragenlijsten is het echter mogelijk dat een verhoogde score niet zozeer de te meten problematiek, maar veeleer de symptomen van de ziekte weergeeft. Een kind met een chronische ziekte wordt in dat geval onjuist gediagnosticeerd. Het is daarom belangrijk een gezondheidsgerelateerde kwaliteit van leven lijst te ontwikkelen speciaal voor kinderen met een chronische ziekte. Deze lijst dient gebaseerd te zijn op de ervaringen van deze groep.

Gezondheidsgerelateerde kwaliteit van leven, de definitie en het meten van kwaliteit van leven bij kinderen

In hoofdstuk 3 worden de definitie en het meten van gezondheidsgerelateerde kwaliteit van leven bij kinderen met een chronische ziekte beschreven. Kwaliteit van leven moet ons inziens onderscheiden worden van de begrippen gezondheid en functionele status, juist door de persoonlijke beleving die bij het beoordelen van kwaliteit van leven hoort. Voorts wordt kwaliteit van leven als een multidimensioneel construct gezien dat het fysiek, sociaal en psychologisch functioneren van personen omvat. De in dit proefschrift gehanteerde definitie van kwaliteit van leven is afgeleid van Calman (1984), die als een van de eersten kwaliteit van leven omschreef als het verschil tussen de verwachtingen (hoop) en het feitelijk functioneren. Gezondheidsgerelateerde kwaliteit van leven wordt in dit proefschrift gedefinieerd als het fysiek, sociaal en psychologisch functioneren van kinderen en hun beleving ten aanzien van problemen in hun functioneren. Het functioneren van kinderen is beschreven als de frequentie van activiteiten, taken, symptomen of problemen en de mate waarin activiteiten goed worden uitgevoerd. Het verschil in functioneren en verwachtingen is gedefinieerd als de beleving van problemen in het fysiek, sociaal en psychologisch functioneren.

Kwaliteit van leven metingen kunnen onder andere gebruikt worden om kinderen te identificeren die extra begeleiding behoeven, om problemen te identificeren van bepaalde groepen en daar normen voor vast te stellen en om de invloed van ziekte en behandeling te evalueren. Kwaliteit van leven metingen kunnen symptomen of beperkingen naar voren halen, die belangrijk zijn maar waar niet automatisch naar gevraagd wordt. Ze kunnen daarmee de arts een handvat bieden in een gesprek met kind of ouder.

Kwaliteit van leven instrumenten kunnen generiek (algemeen) of ziektespecifiek van aard zijn. De generieke instrumenten zijn geschikt voor alle kinderen met een chronische ziekte. Met behulp van deze instrumenten is een vergelijking tussen verschillende ziektebeelden maar, waar relevant, ook een vergelijking met gezonde kinderen mogelijk. De ziektespecifieke instrumenten zijn geschikt voor groepen kinderen met een bepaald ziektebeeld.

Als kwaliteit van leven een subjectief begrip is, dan is het eigen oordeel van het kind belangrijk. Hoewel in onderzoek onder kinderen nog dikwijls wordt gevraagd naar de mening van de ouders, is ook bekend dat het oordeel van ouders en kinderen zeker op minder waarneembare gebieden van elkaar verschilt. Gezien het subjectieve karakter van kwaliteit van leven, zijn wij van mening dat het kind zelf, indien mogelijk, een oordeel moet geven. Dit stelt echter eisen aan het instrument daar het aangepast moet zijn aan het lees- en begripsniveau van de kinderen, in dit proefschrift kinderen van 7 tot en met 13 jaar, de aandacht van hen vast moet houden en slechts een beperkt tijdsbestek kan bestrijken. Het oordeel van de ouders, op dezelfde

manier gevraagd, kan naast het oordeel van het kind worden gelegd. Daar het moeilijk is voor ouders om de beleving van hun kind in te schatten, is het zinnvoller de beleving van de ouders zelf te vragen.

Uit het bovenstaande kunnen de volgende criteria voor het ontwikkelen van kwaliteit van leven vragenlijsten voor kinderen worden afgeleid:

1. De vragenlijst is multidimensioneel, omvat het fysiek, sociaal en psychologisch functioneren en geeft een profiel van het functioneren.
2. De vragenlijst omvat het functioneren en de gevoelens met betrekking tot beperkingen en de problemen in functioneren.
3. De vragenlijst omvat positieve en negatieve emoties.
4. De vragenlijst omvat algemene activiteiten, taken en problemen en ziektespecifieke problemen en behandelingskenmerken.
5. De vergelijkingsgroep omvat zowel kinderen met een chronische ziekte als gezonde kinderen.
6. De vragenlijst vraagt ouders en kinderen naar hun mening over het functioneren van het kind. Gevoelens kunnen echter alleen via zelfrapportage verkregen worden. Kinderen en ouders rapporteren daarom hun eigen gevoelens.
7. De vragenlijst is gemakkelijk voor de kinderen te beantwoorden. De manier van vragen, de woorden die gebruikt worden en de lay-out zijn geschikt voor kinderen uit de doelgroep.

Beschrijving van de HOE GAAT HET? (HOW ARE YOU? HAY)

Geen van de reeds bestaande kwaliteit van leven vragenlijsten bleek te voldoen aan al deze criteria. Besloten werd daarom de HOE GAAT HET? (HAY) te ontwikkelen, een gezondheidsgerelateerde kwaliteit van leven vragenlijst voor kinderen van 7 tot en met 13 jaar met een chronische ziekte. De ontwikkeling van de lijst bestond uit drie fases. Tijdens de eerste fase is de definitie van kwaliteit van leven vastgesteld. Op basis van literatuur en interviews met kinderen, ouders en deskundigen zijn voorlopige items en antwoordcategorieën geformuleerd. Een panel van deskundigen heeft de eerste versie van de HAY beoordeeld. De validiteit en betrouwbaarheid van deze versie zijn in een pilot-studie onderzocht.

De pilot-versie van de HAY bestaat uit een generiek deel dat ook te beantwoorden is door gezonde kinderen, een deel bestemd voor alle kinderen met een chronische ziekte en ziektespecifieke delen voor kinderen met astma, diabetes mellitus, epilepsie en reuma. Het generieke deel omvat de dimensies fysieke activiteiten, cognitieve taken, sociale activiteiten, sociale problemen, fysieke klachten en behandeling. Het deel alleen geschikt voor kinderen met een chronische ziekte omvat zorgen met betrekking tot de ziekte en minderwaardigheidsgevoelens gerelateerd aan het hebben van een ziekte. De ziektespecifieke delen omvat specifieke fysieke klachten en zelfzorg met betrekking tot respectievelijk astma, diabetes mellitus, epilepsie en reuma. Al deze dimensies omvatten vragen naar het voorkomen van bepaalde

activiteiten of problemen, de zogenaamde prevalentievragen. Een voorbeeld is "Heb je de laatste dagen gefietst?". Daarnaast zijn er vragen naar de beleving van kinderen ("Hoe voel je je als fietsen niet goed gaat?"). Deze laatste vragen hebben als antwoordcategorie zeven gezichtjes van verdrietig tot blij. De dimensies fysieke en sociale activiteiten en cognitieve taken omvatten daarnaast ook een vraag naar de kwaliteit van de uitvoering ("Fietsen gaat:"). De dimensies zorgen en gevoelens van minderwaardigheid omvatten alleen een prevalentievraag ("Ben je de laatste dagen blij geweest?").

Resultaten van de pilot-studie

In hoofdstuk 4 worden de resultaten van de pilot-studie naar de bruikbaarheid, validiteit en betrouwbaarheid van de lijst beschreven. Deze studie is verricht onder 89 kinderen met een chronische ziekte en 134 gezonde kinderen en hun ouders. De kinderen met een chronische ziekte en hun ouders zijn allen door een ervaren interviewer thuis bezocht. De gezonde kinderen vulden de vragenlijst in de klas in en ouders vulden de vragenlijst thuis in. Alle ouders en kinderen zijn gevraagd aanvullingen en veranderingen aan te geven.

De pilot-studie wijst uit dat de kinderen de vragenlijst in gemiddeld 30 minuten invullen. De opmerkingen van ouders en kinderen geven echter aan dat enkele vragen en antwoordcategorieën zouden moeten worden aangepast. De vraag naar de beleving van problemen, "Hoe voel je je als ...", heeft zeven antwoordcategorieën met een gezichtje van erg verdrietig tot erg blij. Sommige kinderen vonden het moeilijk om hierop te antwoorden daar zij een andere emotie dan verdrietig of blij voelden, bijvoorbeeld boos. Tevens was het voor kinderen moeilijk een onderscheid te maken tussen de drie positieve gezichtjes.

De validiteit en betrouwbaarheid van de HAY zijn getoetst middels confirmatieve factoranalyse met behulp van LISREL, correlaties tussen schalen van de HAY, Cronbach's alfa's, de correlatie tussen schalen van de HAY en Gedragsvragenlijst voor kinderen (CBCL) (Verhulst, Koot, Akkerhuis & Veerman, 1990) en het toetsen van hypothesen met betrekking tot verschillen in functioneren van kinderen met een chronische ziekte, en de verschillende groepen daarbinnen, en gezonde kinderen. Confirmatieve factoranalyse is uitgevoerd op de vragen naar beleving en emoties van de kinderversie. Op basis van de resultaten zijn de bijpassende schalen naar de prevalentie en de mate waarin kinderen de activiteiten en taken goed konden uitvoeren gevormd. De ouderschalen zijn gevormd op basis van de resultaten van de kinderschalen.

Uit de confirmatieve factoranalyse blijkt een acceptabele fit en goede betrouwbaarheden voor de afzonderlijke schalen met Cronbach's alfa's die variëren van .70 tot .81 in de kinderversie. De ouderschalen hebben een goede betrouwbaarheid met Cronbach's alfa's van .75 tot .90. De vragen naar emoties met

betrekking tot de ziekte, het deel voor alle kinderen met een chronische ziekte, konden echter niet worden geanalyseerd vanwege scheef verdeelde items. De ziektespecifieke delen hebben een redelijke tot goede betrouwbaarheid. De groepen waren echter te klein om middels confirmatieve factoranalyse de structuur te toetsen.

Correlaties tussen de schalen van de HAY en overeenkomstige schalen van de CBCL variëren van .30 tot .51. Hieruit blijkt dat de schalen enige samenhang hebben, maar tevens voldoende unieke variantie hebben om naast elkaar te worden gebruikt.

Vergelijkingen tussen de groep kinderen met een chronische ziekte en gezonde kinderen geven aan dat de meeste hypothesen werden bevestigd. Kinderen met een chronische ziekte hebben vergeleken met gezonde kinderen, volgens hun eigen rapportage, minder fysieke activiteiten ($p < .001$), meer fysieke klachten ($p < .01$), doen minder cognitieve taken ($p < .001$), en hebben meer problemen in de uitvoering van fysieke activiteiten ($p < .001$). Gezonde kinderen daarentegen hebben een negatievere beleving ten aanzien van algemene behandeling dan kinderen met een chronische ziekte ($p < .01$). Volgens de rapportage van de ouders hebben kinderen met een chronische ziekte minder fysieke activiteiten en meer problemen in de uitvoering van fysieke activiteiten ($p < .001$), minder cognitieve taken ($p < .001$), minder goede uitvoering van cognitieve taken ($p < .01$), minder sociale activiteiten ($p < .01$) en meer fysieke klachten ($p < .001$). Bovendien hebben ouders van kinderen met een chronische ziekte een negatievere beleving ten aanzien van problemen in fysieke activiteiten ($p < .01$), sociale problemen ($p < .001$) en fysieke klachten ($p < .001$) dan ouders van gezonde kinderen.

De correlaties tussen de ouder- en kindschalen, variërend van .15 tot .57, geven aan dat de lijsten aanvullend op elkaar dienen te worden gebruikt.

Op grond van de resultaten in de pilot-studie zijn de volgende verbeteringen in de HAY aangebracht:

1. Het algemene deel omvat de dimensies fysieke activiteiten, cognitieve taken, sociale activiteiten, sociale problemen, fysieke klachten, behandeling, positieve gevoelens en negatieve gevoelens. Hieraan zijn de dimensies positieve en negatieve gevoelens toegevoegd.
2. De dichotome antwoordcategorieën bij de prevalentievragen zijn uitgebreid tot vier antwoordcategorieën (van "Nee nooit" tot "Ja, heel vaak").
3. De vraag naar de beleving van problemen zijn geherformuleerd naar "Hoe vind je het als fietsen niet goed gaat?".
4. De antwoordcategorieën van de vragen naar de beleving van problemen of beperkingen zijn één positief gezichtje en drie gezichtjes van neutraal tot erg verdrietig met als onderschrift "niet erg" tot "heel erg".
5. De inhoud van de ziektespecifieke modules is aangepast. Alle ziektespecifieke delen omvatten specifieke fysieke klachten en zelfzorg.

De validiteit en betrouwbaarheid van de HAY

In hoofdstuk 5 zijn de resultaten van de hoofdstudie beschreven waarin de validiteit en betrouwbaarheid van de HAY zijn getoetst onder een grote groep kinderen met een chronische ziekte ($n=577$) en een groep gezonde kinderen ($n=418$).

In de hoofdstudie zijn de ouders en kinderen benaderd via hun arts of school middels een brief. De ouders konden zichzelf en hun kind aanmelden via een antwoordstrookje. Alle kinderen en ouders hebben de lijst thuis ingevuld. In de begeleidende brief werd gevraagd de lijst apart van elkaar in te vullen. De kinderen ontvingen een klein cadeautje voor hun deelname, de ouders een kort verslag.

Constructvaliditeit is getoetst middels confirmatieve factoranalyse waarbij gebruik werd gemaakt van LISREL. De confirmatieve factoranalyse is gedaan op de vragen naar de beleving en emoties van kinderen. De hieruit gevonden structuur is opgelegd aan de overige items, zodat schalen naar de prevalentie, de kwaliteit van uitvoering en de schalen naar de beleving van problemen dezelfde items bevatten. De ouderlijst is gebaseerd op de uitkomsten van de kinderlijst. Multisample analyse is uitgevoerd om te bepalen of de gevonden structuur in alle groepen stabiel was. Voorts is de relatie tussen schalen van de HAY en bestaande andere instrumenten, de Gedragsvragenlijst voor kinderen (CBCL) (Verhulst, Koot, Akkerhuis & Veerman, 1990) en een vertaalde en gewijzigde versie van de Child Attitude Toward Illness Scale (CATIS) (Austin & Huberty, 1993) onderzocht.

Vervolgens zijn de hypothesen met betrekking tot de verschillen tussen groepen getoetst met een twee-factoriële variantie-analyse middels GLM (SPSS) met sekse en ziekte als factoren en leeftijd als covariaat. Hiertoe werden de volgende hypothesen opgesteld. Van kinderen met astma werd verwacht dat zij in vergelijking met gezonde kinderen meer fysieke klachten zouden hebben, meer fysieke belemmeringen en meer belemmeringen in sociale activiteiten alsmede een hoger schoolverzuim. Kinderen met diabetes mellitus zouden meer problemen hebben in sociaal functioneren dan gezonde kinderen. Kinderen met epilepsie zouden meer problemen hebben met cognitieve taken en sociaal functioneren dan gezonde kinderen. Kinderen met reuma zouden meer fysieke klachten, meer problemen in hun fysieke en sociale activiteiten en een hoger schoolverzuim hebben dan gezonde kinderen. Er werden geen hypothesen geformuleerd over verschillen in gevoelens of waarderingen van problemen, daar deze beïnvloed kunnen worden door vele variabelen, zoals de doelen en waarden die een kind heeft. Tot slot werden de relaties tussen ouder en kind rapportage middels intraclass correlations (ICC) onderzocht (Deyo, Diehr & Patrick, 1991).

De onderzoeksgroep bestond uit 577 kinderen (291 jongens, 286 meisjes) met een chronische ziekte (275 kinderen met astma, 139 kinderen met diabetes mellitus, 104 kinderen met epilepsie en 59 kinderen met reuma) en 418 gezonde kinderen (201

jongens, 217 meisjes). De kinderen met een chronische ziekte en gezonde kinderen hadden een gemiddelde leeftijd van 10 jaar. De inclusiecriteria voor de kinderen met een chronische ziekte waren dat zij tenminste één keer het afgelopen jaar een arts hadden bezocht in verband met hun chronische ziekte en dat zij maar één chronische ziekte hadden. Voorts bezochten alle kinderen een normale basisschool en waren zij, maar ook hun ouders, in staat om Nederlands te lezen. Er zijn geen statistisch significante verschillen tussen beide groepen in sekse, leeftijd, nationaliteit, scholing ouders, het percentage één- of twee-oudergezinnen en het aantal kinderen in een gezin.

Confirmatieve factoranalyse en betrouwbaarheidsanalyses geven aan dat het generieke deel van de HAY bestaat uit 5 dimensies met een goede fit en goede betrouwbaarheden: fysieke activiteiten, cognitieve taken, sociale activiteiten, fysieke klachten en positieve emoties. Multisample analyse geeft aan dat de gevonden structuur in alle groepen stabiel is. De Cronbach's alfa's in de kinderlijst variëren van .77 tot .86 en in de ouderlijst van .86 tot .93. Het deel voor kinderen met een chronische ziekte bevat twee dimensies met een goede fit en goede betrouwbaarheden, namelijk zorgen en gevoelens van minderwaardigheid gerelateerd aan het hebben van een chronische ziekte. De Cronbach's alfa's van deze twee dimensies in de kinderlijst zijn respectievelijk .80 en .74, en .86 en .66 in de ouderlijst. Multisample analyses geven aan dat de structuur stabiel is in alle groepen.

De ziektespecifieke dimensies zijn voor elke groep verschillend. Het astmaspecifieke deel bestaat uit twee dimensies met een goede fit en goede betrouwbaarheden, namelijk fysieke klachten en zelfzorg. De Cronbach's alfa's in de kinderlijst zijn respectievelijk .77 en .76, en .83 en .78 in de ouderlijst. De diabetesspecifieke lijst bestaat uit drie dimensies, te weten fysieke klachten gerelateerd aan een hyper, fysieke klachten gerelateerd aan een hypo en zelfzorg. Hoewel het model een goede fit heeft, zijn de Cronbach's alfa's laag, respectievelijk .58, .63 en .63 in de kinderlijst en .62, .67 en .71 in de ouderlijst. Het epilepsiespecifieke deel bestaat uit drie dimensies met een goede fit en een goede betrouwbaarheid, namelijk fysieke klachten, aanvallen en zelfzorg. De Cronbach's alfa's zijn respectievelijk .74, .78 en .70 in de kinderlijst en .72, .83 en .76 in de ouderlijst. Het reumaspecifieke deel bestaat eveneens uit drie dimensies en bestaat uit fysieke klachten, beperkingen en zelfzorg. De Cronbach's alfa's van deze dimensies zijn respectievelijk .73, .74 en .76 in de kinderlijst en .81, .73 en .74 in de ouderlijst. De resultaten van de confirmatieve factoranalyse waren echter niet stabiel, mogelijk in verband met het relatief kleine aantal kinderen in deze laatste groep.

De correlaties tussen de schalen "zich zorgen maken over de ziekte" en "gevoelens van minderwaardigheid" en de herziene versie van de CATIS zijn .53 en .59. Hieruit blijkt dat de schalen enige samenhang hebben, maar tevens voldoende unieke variantie hebben om naast elkaar te worden gebruikt.

De hoogste correlaties tussen schalen van de HAY en de CBCL zijn gevonden op cognitieve competentie (.58) en fysieke klachten (-.47). De correlaties op fysieke competentie (.16) en sociale competentie (.26) zijn laag. Een reden hiervan zou kunnen zijn dat de fysieke en sociale competentie schalen in de CBCL tevens de deelname van het kind aan activiteiten meten, terwijl de schalen van de HAY die hier gebruikt werden alleen competentie, de kwaliteit van uitvoering, meten.

De hypothesen met betrekking tot de verschillen in groepen zijn gedeeltelijk bevestigd. Kinderen met een chronische ziekte hebben vergeleken met gezonde kinderen significant minder sociale activiteiten ($p < .01$), meer problemen in de uitvoering van fysieke activiteiten ($p < .01$) en meer negatieve gevoelens ten aanzien van problemen in sociale activiteiten ($p < .01$) volgens hun eigen rapportage. In tegenstelling tot wat verwacht werd, rapporteren de kinderen met een chronische ziekte niet meer algemene fysieke klachten dan gezonde kinderen. Een verklaring hiervoor is moeilijk te vinden. Mogelijk zijn voor kinderen met een chronische ziekte de specifieke klachten meer van belang en worden algemene klachten minder opgemerkt. Tevens zijn er geen significante verschillen tussen de beide groepen ten aanzien van de prevalentie van fysieke activiteiten, cognitieve taken, de uitvoering van sociale activiteiten en cognitieve taken.

Volgens hun ouders hebben kinderen met een chronische ziekte vergeleken met gezonde kinderen significant minder sociale activiteiten ($p < .01$), meer algemene fysieke klachten ($p < .01$) en meer problemen in de uitvoering van fysieke activiteiten, cognitieve taken en sociale activiteiten ($p < .01$). Ouders van kinderen met een chronische ziekte hebben meer negatieve gevoelens over beperkingen in de sociale activiteiten van hun kind dan ouders van gezonde kinderen ($p < .01$). Ouders van kinderen met een chronische ziekte rapporteren echter geen lagere prevalentie van fysieke activiteiten of cognitieve taken van hun kind. Een mogelijke verklaring hiervoor kan zijn dat juist de kwaliteit van uitvoering, meer dan de prevalentie, wordt beperkt bij kinderen met een chronische ziekte zoals de ziekten bestudeerd in dit proefschrift.

Gemiddeld rapporteren meisjes (en hun ouders) significant minder fysieke activiteiten en meer fysieke klachten dan jongens. Tevens hebben de meisjes significant meer negatieve gevoelens ten aanzien van fysieke klachten dan jongens ($p < .01$). Er zijn geen significante interactie-effecten tussen sekse en het hebben van een ziekte.

Met betrekking tot de verschillen tussen de vier groepen kinderen met een chronische ziekte en de gezonde kinderen, zijn de volgende resultaten gevonden. Zoals verwacht rapporteren kinderen met astma significant meer problemen in de uitvoering van fysieke activiteiten ($p < .01$) dan gezonde kinderen volgens zowel kind- als ouder rapportage. Ouders rapporteren ook significant meer fysieke klachten van hun kinderen dan ouders van gezonde kinderen ($p < .01$). Kinderen met astma en hun

ouders rapporteren significant minder sociale activiteiten en meer problemen in de uitvoering van sociale activiteiten ($p < .01$) dan gezonde kinderen en hun ouders. De kinderen met astma rapporteren zelf significant meer negatieve gevoelens ten aanzien van beperkingen in sociale activiteiten ($p < .01$). Deze resultaten geven aan dat sociale activiteiten in onderzoek en bij de begeleiding van kinderen met astma betrokken dienen te worden.

De resultaten voor kinderen met diabetes mellitus gevonden in dit onderzoek komen overeen met de conclusies van de studies die in hoofdstuk 2 beschreven zijn. Kinderen met diabetes mellitus scoren net zo hoog of hoger dan gezonde kinderen. Kinderen met diabetes mellitus hebben zelfs significant meer fysieke activiteiten dan gezonde kinderen volgens kind- en ouderrapportage ($p < .01$). In tegenstelling tot wat verwacht werd, hebben kinderen met diabetes mellitus niet meer problemen in sociale activiteiten dan gezonde kinderen. Meer onderzoek hiernaar is, gezien het aantal studies waar de hypothese op was gebaseerd, echter wenselijk. Ouders van kinderen met diabetes mellitus rapporteren meer fysieke klachten van hun kind dan ouders van gezonde kinderen ($p < .01$). Mogelijk wordt dit mede veroorzaakt door het feit dat hypo's en hyper's samengaan met een aantal van de klachten die tot algemene klachten horen, zoals hoofdpijn of misselijkheid. Of ouders deze klachten overrapporteren of kinderen onderrapporteren is niet bekend. In de klinische praktijk zal men alert moeten zijn op deze klachten en de waarneming daarvan door ouders en kinderen.

Zoals verwacht hebben kinderen met epilepsie volgens de ouders meer beperkingen in het cognitief functioneren dan gezonde kinderen. Kinderen rapporteren echter geen problemen in cognitief functioneren. Het is mogelijk dat kinderen deze problemen onderrapporteren. Het is anderzijds ook bekend dat de mening van ouders mogelijk beïnvloed wordt door de lagere verwachtingen die zij van hun kind hebben (Aldenkamp et al., 1993). Kinderen met epilepsie noch hun ouders scoren meer problemen in sociale activiteiten. Hierbij moet worden opgemerkt dat het aantal studies waarop de hypothese gebaseerd was, klein was. Sociale activiteiten zullen in onderzoek maar ook in de klinische praktijk, meer onderzocht moeten worden.

Kinderen met reuma hebben zoals verwacht significant meer problemen in de uitvoering van fysieke activiteiten dan gezonde kinderen volgens zowel kinder- als ouderrapportage. Bovendien hebben zowel kinderen als hun ouders meer negatieve gevoelens ten aanzien van beperkingen in fysieke activiteiten. Daarnaast hebben ouders en kinderen meer negatieve gevoelens ten aanzien van beperkingen in sociale activiteiten dan gezonde kinderen en hun ouders ($p < .01$). Een mogelijke verklaring voor het niet vinden van significante verschillen in de prevalentie van fysieke klachten is dat bij reuma de specifieke klachten meer op de voorgrond staan. Dat er geen verschillen zijn gevonden in sociale activiteiten is moeilijk te verklaren, zeker gegeven

het feit dat sociale activiteiten voor een deel ook fysieke activiteiten zullen betreffen in deze leeftijdsgroep. Verder onderzoek hiernaar is wenselijk.

Zoals al eerder werd aangegeven, is het van belang het functioneren van een bepaald persoon te relateren aan de gevoelens met betrekking tot problemen in functioneren. Wanneer kinderen klachten of beperkingen ervaren, en deze als belastend ervaren, dan zullen deze als onderwerp van behandeling moeten dienen ofwel in de behandeling van symptomen ofwel in het veranderen van verwachtingen.

Het vermogen van de HAY om binnen een bepaalde groep te differentiëren afhankelijk van de ernst van de ziekte is getoetst door de mate van ziekteklachten (door ouders gerapporteerd) te relateren aan het functioneren van het kind, na gecontroleerd te hebben voor leeftijd en sekse. Met name voor kinderen met astma en reuma blijkt dat de mate waarin zij ziekteklachten hebben, gerelateerd is aan beperkingen in hun functioneren zoals gerapporteerd in de HAY. Een mogelijke verklaring voor het niet vinden van deze gevoeligheid voor ziekteklachten bij de kinderen met diabetes en epilepsie, is dat het moeilijker was uit de beschikbare data een goede maat voor de ernst van de klachten van deze kinderen te formuleren. Het toetsen van de gevoeligheid voor veranderingen was in deze studie nog niet mogelijk. Voor de bepaling hiervan in de toekomst wordt aangeraden gegevens gerelateerd aan verandering in kwaliteit van leven te relateren aan "objectieve maten" van ernst binnen een bepaalde groep.

Tot slot is de overeenkomst tussen kind- en ouder rapportage onderzocht. In de groep gezonde kinderen zijn de intraclasscorrelaties tussen ouders en kinderen hoger voor de prevalentieschalen dan voor de schalen die de kwaliteit van de uitvoering van activiteiten bepalen. Een mogelijke verklaring hiervoor kan zijn dat de prevalentie van activiteiten beter door de ouders kan worden waargenomen dan de kwaliteit van uitvoering en minder gevoelig is voor subjectieve oordelen van kind of ouder. Daarnaast zijn bij kinderen met een chronische ziekte de correlaties tussen kinderen en ouders op de schalen die de kwaliteit van uitvoering meten hoger dan bij de gezonde kinderen en hun ouders. Een mogelijke verklaring hiervoor is dat ouders van kinderen met een chronische ziekte meer op het functioneren van hun kinderen letten dan ouders van gezonde kinderen. Met name voor ouders van kinderen met een chronische ziekte geldt dat zij meer beperkingen in fysieke, cognitieve en sociale activiteiten en minder blijde emoties van hun kinderen aangeven dan de kinderen zelf. In tegenstelling hiermee rapporteren zij echter ook minder fysieke klachten dan hun kinderen. Deze resultaten zouden kunnen leiden tot de hypothese dat ouders een meer pessimistische kijk hebben op hoe hun kinderen functioneren dan hun kinderen zelf. Daarnaast laten deze resultaten zien dat interne processen, zoals fysieke klachten, mogelijk minder goed door ouders worden waargenomen. Het wordt aanbevolen beide rapportages naast elkaar te gebruiken.

Discussie

In hoofdstuk 6 volgt de algemene discussie en worden aanbevelingen voor verder onderzoek gedaan. De volgende eigenschappen van de HAY worden besproken: multidimensionaliteit, geschiktheid voor de leeftijdsgroep, gebruik van vragen naar de beleving, inclusie van positieve en negatieve emoties, gebruik van generieke en ziektespecifieke delen, het samengaan van kind- en ouderreportage. Tevens wordt ingegaan op de methode van onderzoek.

Aanbevelingen

Naar aanleiding van dit onderzoek worden de volgende aanbevelingen gedaan:

1. De validiteit en betrouwbaarheid van de ziektespecifieke delen van de HAY voor kinderen met diabetes mellitus dient nader onderzocht te worden.
2. De validiteit en betrouwbaarheid van de HAY voor kinderen met reuma dient in een grotere groep geconfirmeerd te worden.
3. Ziektespecifieke modules voor kinderen met een andere ziekte dan astma, diabetes mellitus, epilepsie en reuma dienen te worden ontwikkeld. Tijdens de ontwikkeling van de HAY zijn er ziektespecifieke modules voor kinderen met nierziekte en kanker ontwikkeld, maar van deze modules zijn de validiteit en betrouwbaarheid nog niet vastgesteld.
4. De gevoeligheid van de HAY voor veranderingen over tijd en in het ziektebeeld dient te worden vastgesteld. Bij voorkeur dient hier gebruik te worden gemaakt van een objectieve maat van ernst.
5. Versies voor kinderen ouder dan 14 jaar en jonger dan 7 jaar dienen te worden ontwikkeld.
6. Slechts weinig kinderen met een niet-Nederlandse achtergrond hebben aan het onderzoek meegewerkt. Het gebruik van de HAY dient ook in deze groep te worden getoetst.
7. Hoewel 20% van de kinderen een chronische ziekte heeft, zijn er vele ziekten waarvan de prevalentie laag is. Juist in deze groepen is het van belang om *internationaal onderzoek te verrichten en resultaten van verschillende landen met elkaar te vergelijken*. Het gebruik van de HAY in andere landen en culturen dient nader te worden onderzocht.
8. Verder onderzoek naar het gebruik van de CATIS in Nederland is zinvol. Daarnaast dient het gebruik van de CBCL onder kinderen met een chronische ziekte nader te worden onderzocht.
9. Meer onderzoek is nodig naar de *predictieve variabelen van gezondheidsgerelateerde kwaliteit van leven van kinderen*, waarbij gebruik dient te worden gemaakt van een theoretisch model en een totaalbeeld van het functioneren van het kind over meerdere dimensies. Alleen dan komt er meer informatie beschikbaar over hoe negatieve consequenties van ziektes voor kinderen zoveel mogelijk voorkomen kunnen worden.

Conclusie

Samenvattend kan gesteld worden dat gezondheidsgerelateerde kwaliteit van leven in de kindergeneeskunde een belangrijk onderwerp is en dat er steeds meer mogelijkheden zijn om gezondheidsgerelateerde kwaliteit van leven van kinderen te bepalen. Een voorbeeld hiervan is de HAY, een valide en betrouwbaar instrument voor kinderen met een chronische ziekte van 7 tot en met 13 jaar. In de klinische praktijk kunnen kwaliteit van leven bepalingen gebruikt worden om de communicatie tussen de arts, kinderen en ouders te verbeteren. In onderzoek kunnen kwaliteit van leven bepalingen gebruikt worden bij de beschrijving van de problemen bij een bepaalde groep en het evalueren van interventies. De aanbevelingen die hierboven gedaan zijn, zullen moeten leiden tot een verdere ontwikkeling van gezondheidsgerelateerde kwaliteit van leven metingen en een beter gebruik van gezondheidsgerelateerde kwaliteit van leven bepalingen in zowel onderzoek als de klinische praktijk.

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APPENDIX 1 - TABLES

- Table 2.1** Clinical studies on the functioning of children with asthma
- Table 2.2** Clinical studies on the functioning of children with diabetes mellitus
- Table 2.3** Clinical studies among children with epilepsy
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- Table 3.1** Health status and quality of life instruments for children – an overview
- Table 5.6** Medical care
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Table 2.1 Clinical studies on the functioning of children with asthma

Authors	Group	N	Age	Measures	Results
Bremberg, 1985	asthma	329	7-15	questionnaire on asthma and health-p	Only first authors are mentioned. c=child report, p=parent report, t=teacher report, ph=physician report 210 stated symptoms, 21% no restriction, 37% 1-9 days restriction, 39% 10-99 days restriction, 100 days or more 3% restriction during the past year 43% used daily drugs mean time absence of school 3 days/year 78% during physical education restricted, during arts & crafts 12%, other lessons 11% school districts were ranked with respect to absence, restriction, perceived understanding of asthma and dustiness
Viney, 1985	asthma healthy	53 42	4-12	interview-content analysis-c	no evidence of suppressed emotional reactions among asthma children with asthma more anger and more feelings of competence than controls preadolescent more depression and fewer good feelings than the younger
Suess, 1986	healthy asthma	90 30	9-18	Brenton Visual Retention Test-c Wechsler Memory Scale-c	steroid theophylline medication regimens, 6-8 hours after medication, can negatively affect cognitive performance children using theophylline alone did not differ from non-asthmatics on these tasks time dependent effects on visual retention and verbal learning were reversible at later times prolonged administration may affect classroom performance both in short run and longitudinally
Marx, 1986	asthma healthy	18 18	7-12	Heart Rate Facial Action Coding System	no deficit in facial expression was found in asthma group to the contrary, asthma more expressions of anger/aggression and emotion expression in total in child-alone condition mothers of asthmatic children showed more happiness hear rate didn't differentiate during any phase of experiment
Hazzard, 1986	asthma attended camp did not attend	43 37	7-15	knowledge scale-c Children health locus of control-c Piers-Harris Children's Self-Concept Scale-c Parent questionnaire-p	children were well informed about condition children tended to have an internal locus of control children self concept higher than mean for standardization healthy sample increasing age and education children made more knowledgeable and more able to control their own health status asthmatic children were fairly active, exercising at least several times a week more asthma related behavior problems in more severe asthma children on camp did not differ significantly from not camp on psychosocial measures

Table 2.1
Clinical studies on the functioning of children with asthma

Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report					
Authors	Group	N	Age	Measures	Results
Chiari, 1987	boys asthma boys healthy	20 20	9-11	Interview (PL Harris)-c	concerning anger: healthy boys see themselves as capable of exerting control, asthma only change of direction of mental problems concerning fear: emotional state can be modified by changing direction to mental processes
Eiser, 1988	asthma healthy	49 49	7-16	General and asthma knowledge questionnaire-c	compared with a healthy group, asthmatic children less well informed about general knowledge of the body knowledge on asthma was also poor, few children avoided specific situations no significant increase with age in knowledge of allergens or awareness of how to control attacks
Perrin, 1989	asthma	46	5-16	Health Resources Inventory-p asthma severity by clinical history severity by parent's judgment-p	parental estimates of severity were significantly correlated with objective measures, although concordance value of 54% indicating a marked lack of agreement between parents and objective ratings children with asthma as a group normal adjustment scores mild or severe asthma had significantly lower adjustment scores than moderate according to parents' ratings objective severity was not associated with adjustment scores, although medication use was significantly associated with lower adjustment scores all levels of severity may be associated with adjustment problems
Furrow, 1989	inpatient asthma	48	6-16	Child Behavior Checklist-p	while problems were generally elevated with non-referred children, most pronounced effects were found for internalizing as opposed to externalizing behaviors. boys scoring behavior problems equal to those of clinically referred a higher frequency of somatic complaints than norm among young asthmatics (6-11)
Hambley, 1989	asthma	54	5-16	Family adaptability and cohesion evaluation scale- FACES III-p Child Behavior Checklist-p Coopersmith Self Esteem Inventory-c Nowicki Strickland Locus of Control Scale for Children-c	children presented with moderate to severe asthma mostly diagnosed before 3 years and often associated with atopic dermatitis families exhibit normal levels of bonding and flexibility in response to stress most children were experiencing behavioral and school-related problems with 6-11 year old boys exhibiting global social competency problems as well girls lower self-esteem than boys locus of control within normal range for all age groups

Table 2.1 Clinical studies on the functioning of children with asthma						
Authors	Group	N	Age	Measures	Results	Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report
Gutstadt, 1989	asthma (severe)	99	9-17	Woodcock Reading mastery Test-c Key Math Diagnostic Arithmetic Test Woodcock-Johnson-c PsychoEducational Battery-c Stossom Intelligence Test-c Child Behavior Checklist -p	academic abilities were average or above average even though children were absent from school one fifth of the days in semester prior to testing low socioeconomic status, older age, history of continuous oral steroid use and presence of emotional and behavioral problems were associated significantly with low performance scores	
Weston, 1989	population: nonasthmatic (78%) asthmatics (16%) asthma in remission (4%)	408	11-13	questionnaire in exercise and sport-c Sport Competition Anxiety Test-c Self concept in physical domain from Harter (adjusted)-c questionnaire on enjoyment in sport-c motivation questionnaire (adjusted)-c	children with higher frequency of asthma enjoyed fitness less and felt that asthma deterred them from doing more exercise, but there was no effect on reported frequency children with asthma were more frequently active and more anxious prior to exercise, but had no higher perceived level of activity children with asthma did not differ from controls in enjoyment of exercise or in physical self concept motives for performing sports were similar, although children with asthma received less pressure to be active	
Townsend, 1991	asthma	100	7-18	self constructed questionnaire-c	children cited respiratory symptoms frequently as bothersome, emotion related items cited frequently but were considered as less bothersome 85% of the children cited running as problematic concerning asthma medication, 32% cited difficulty falling asleep, not being able to sit still and restlessness as a consequence of medication 38% responded 'owning a pet' as most important wish	
Groot, 1992	population	217 6 162 1	5-6 10-12	CARA-index-ph	prevalence of asthma 6.9% tot 11.6% younger children more absenteeism than older children (31% versus 12%), younger children visit GP more often (63 versus 44%) younger children use more often medication (23 versus 11) one third of school absenteeism caused by airway problems boys visit GP more often for airway problems boys more asthma complaints than girls humid houses and patients ensured by the Dutch National Health Service more complaints	

Table 2.1 <i>Clinical studies on the functioning of children with asthma</i>					
Authors	Group	N	Age	Measures	Results
MacLean, 1992a	asthma	81	6-14	illness severity pr CBCL-p Life Events Scale-pr	<p>Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report</p> <p>Results somatic complaints higher with exception of boys 12-14 than norms high severity poorer social adjustment boys of 6-11 had a significantly different total social competence scores, with 5% within clinical range total behavior problems significant higher for children with asthma, 29.5 above 90th percentile, 11.5% scores higher than 98th percentile internalizing score was higher (59.6) than externalizing score (55.1) significant relation between age and gender for the school scale, younger girls and older boys significantly higher t-scores negative life events significantly related to total behavior problems and internalizing behavior problems higher severity was significantly correlated with more total behavior problems and total social competence and social competence none of the predictor variables accounted for a significant amount of variance in the externalizing scores higher SES correlated negatively with higher scores on activity and social subscale higher severity correlated with higher social subscale scores negative change correlated with less optimal school performance</p>
Butz, 1993	asthma	155	7-12	State-Trait Anxiety Inventory for Children-c State-Trait Anxiety Inventory form-p questionnaire-p and c	<p>children were followed for an 8-week period mother's state and trait anxiety scores were consistently higher than the child's scores no correlation between child and mother anxiety levels almost two-third of children reported feeling "panic" at the beginning of the asthma attack, which was significantly related with child state anxiety as well as one or less attacks during the last 12 months and 2-10 attacks during the last 12 months child state anxiety level, mild-to-moderate rating of usual attacks reported by the mother, and no panic at the beginning were related to child trait anxiety</p>
McNaughton 1993	asthma	381	6-11	structured interview-c and p role play-c and p ratings on reading achievement and interference in life at school-t	<p>large differences on school attendance; school, economic and cultural variables were related to school attendance, characteristics of family were related to reading achievement and interference in school life effective management is facilitated in conditions of low stress, with the availability of social resources and little need of contact with official agencies</p>

Table 2.1 Clinical studies on the functioning of children with asthma

Authors	Group	N	Age	Measures	Results
Meijer, 1995	I. controlled asthma uncontrolled II controlled asthma uncontrolled	20 20 15 15	9-15	diagnosis controlled asthma, severity and allergy ph Interactional Problem Solving Inventory-p Family Dimension Scale- parents Neuroticism and neurotic- somaticism subscales of the Amsterdam Biographical questionnaire for Children-c	Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report Results cohesion of family members and rigid manner of function of caregivers may have a positive rather than a negative influence on the welfare of the child controlled asthma correct use of medication which was predominantly evident in more structured and independent family environments distinction between controlled and uncontrolled asthma leads to a better insight of psychosomatic variables
Graetz, 1995	asthma healthy	21 21	8-13	Sociometric questionnaire-c Liking Rating Scale-c Revised Class Play with added five roles relevant for asthma-c Loneliness Scale-c	no significant differences on popularity, reciprocated friendships, rejection, self-perceived loneliness between the two groups no significant differences on social reputation for sociability-leadership, aggressive-disruptive and sensitive-isolation no significant differences on social reputation for illness behaviors and sporting ability children with asthma had higher levels of illness-related behaviors frequency with hospitalization was significantly negatively correlated with liking frequency with hospitalization significantly positively correlated with loneliness and sensitive-isolated scale
Eksi, 1995	healthy mild asthma moderate asthma	60 29 31	4-16	Pearlman-Biermann classification of severity Child Behavior Checklist (CBCL)-p semi-structured interview with psychologic and psychiatrist-p	CBCL scores on social, activity, school and total social competence were higher for healthy children, indicating a lower performance of children with asthma CBCL scores on behavior problems were higher (indicating more problems) for asthmatic children girls higher scores on school, total competence and somatic problems, boys higher ratings on aggression, externalizing behaviors and total problem scores problem scores for children with asthma were significantly correlated with parental friction at home, unsatisfactory relationships with siblings and other health problems neither CBCL total problem scores nor social competency were correlated with duration or severity illness clinging, childish behavior, dependence, a demanding nature, and using illness in service of achieving aims were common or widespread in the asthma group not only the children but also other family members experienced problems related to child's illness

Table 2.1 Clinical studies on the functioning of children with asthma

Authors	Group	N	Age	Measures	Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report
Bussing, 1996	asthma healthy	37 31	7-17	interview DSM-III-R anxiety disorder with the Schedule for Affective Disorders and Schizophrenia for School-Age children-c and p Index Family Relations (IFR)-c and p	Results asthma group more anxiety disorders than controls, separation anxiety most common asthma group more past school problems, school refusal and repeated a grade, 25% compared to 6% of the healthy children needed special school placement asthma group more past psychiatric illnesses also family members more psychiatric problems asthma group more interfamilial stress; 24% of the parents, 30% of the children scoring in clinical range

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures
Holmes, 1985	diabetes	42	6-16	WISC-R-c children with reading deficit: Bender Visual Motor Gestalt Test-c Rey Auditory Verbal Learning Test-c Digit Span-c
Gross, 1985	non-diabetes diabetes	30 37	9-12	Piers-Harris Self-concept Scale-c Nowicki-Strickland Internal- External Locus of Control scale-c children's health locus of control scale-c HbA1c
Ryan, 1985	diabetes non-diabetes	125 83	10-19	Wechsler Intelligence Scale for Children (subtests)-c Boston Embedded Fig. Test-c Hooper Visual Organization Test-c Block Design Test-c Standardized Road Map Test-c Symbol Digit Learning Test-c Ryan's verbal learning test-c Delay Verbal Recall Test-c WAIS Digit Symbol Substitution Test-c Wide Range Achievement Test Trail Making Test-c Grooved Pegboard-c Piers-Harris Children's Self- concept Scale-c

Only first authors are mentioned.
c=child report p=parent report, t=teacher, ph=physician report

Results:
significant lower WISC-R Performance IQ for children with early onset (<7 years) and long disease duration (>5 years)
performance subtests lower for this group, slower responding to time tasks may account for lower scores
children with early onset-long duration also higher rates of reading and memory impairment IQ scores nevertheless in average range

no significant correlations between HbA1c and other measures of locus of control and self concept
no significant differences on locus of control or self concept between control group and diabetes diabetes significantly more internal health locus of control

children with early onset (<5 years of age) performed more poorly than adolescent than later onset or nondiabetics on virtually all tests, including measures of intelligence visuospatial ability, memory, motor speed and eye-hand coordination
however all three groups mean scores within "normal" range
24% of the early onset and 6% of later onset feel in "impairment group"
age at onset and duration affect neuropsychological functioning
duration best predicted performance on those tests highly overlearned, primarily verbal skills, age at onset best predicted scores on tests requiring the ability to process relatively unfamiliar typically nonverbal information in novel ways

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Only first authors are mentioned. c = child report p = parent report, t = teacher, ph = physician report
Kovacs 1986	newly diagnosed	74	8-13	Clinical interviews with parents and children separately-c and p Children's Depression Inventory-c Revised Children's Manifest Anxiety Scale (RCMAS)-c Coopersmith's Self-Inventory (SEI)-c CBCL social competence-p Back Depression Inventory-p SCL-90-p	Results: few emotional symptoms and good self-esteem, but symptoms of depression and anxiety decreased and self-esteem increased with time methods of parent and child report not interchangeably clinical assessments and self-assessments tap different constructs children are more comfortable with aspects of diabetic care than previously thought
Johnson, 1986	diabetes	168	6-19	interview-p and c	parent-child agreement was good, youngest children showed less agreement on some measures (for instance time, type of diet), also girls better agreement on three measures adherence appears to be a complex construct teenagers were less adherent on most measures than younger children
Rovet, 1987	early onset diabetes late onset diabetes sibling controls	27 24 30	6-13	Child Behavior Checklist-p Draw a person test-c Piers-Harris Children's Self concept Scale-c Middle Child Temperament Scale-p School performance-p Wide Range Achievement Test- c	children with late onset (>4 years) diabetes boys showed increased incidence of behavior problems, 83% of whom could be diagnosed a diagnostic classification "schizoid" most frequent and more distortions of body image, girls no behavioral subtype classification early onset boys and girls higher total behavior problem score sisters had elevated internalizing scores girls early onset lower school performance and social competence, boys late onset difficulty in social relations results were not related to any indices of diabetic control, background factors early onset girls more hypoglycemic seizures effects are most severe for latency-aged boys
Court, 1988	diabetes healthy	127 51	8-16	HbA1 and c-peptide social information (5 items)-p Rutter's A2 scale -p Rutter's B2- teacher multiple choice questionnaire on knowledge-p and c (11 years and older)	25% of the diabetes mellitus and 14% of the controls were rated by their parents and their teachers to be disturbed no relation between extent of Behavior problems and control of diabetes, age, age at diagnosis, duration children with younger parents tended to be perceived by their parents as more disturbed as were diabetics from families facing social problems the more knowledge families have, the less likely to have disturbed children

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
Rovet, 1988	diabetes: early onset diabetes late onset diabetes sibling controls	51 (27) (24) 30		WISC-R Similarities and Vocabulary Subtests-c SKA Primary Mental Abilities-c Verbal Meaning Test-c WISC-R Block design and object assembly subtests-c PMA Spatial Relation subtest-c Berry-Buktenica Test of Visual Motor Integration-c WideRange AchievementTest-c WAIS-short form-p interview-p	Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report children with early onset, particularly girls, scored lower on tests of visuospatial but not verbal ability than other groups spatial ability and arithmetic skill related to illness factors many children with early onset difficulties at school early onset more hypoglycemic convulsions duration of illness, age of onset and hypoglycemic convulsions predicted spatial ability, but also age was a predictor
Varni, 1989	diabetes: adolescents children	41 34	6-17	Family Relationship Index (FRI) of the family Environment Scales-c Self Perception Profile for Children-c Child Behavior Checklist-p	for children, family social support and peer social support were significant predictors for internalizing and externalizing behavior for adolescents, peer social support significant predictor for internalizing and externalizing behavior problems general self-esteem trend toward significance overall model 54% of the variance for internalizing and 25% for externalizing behavior problems
Roth, 1989	diabetes	40	9-16	Fear of failure questionnaire-c Social insecurity questionnaire-c Diabetes attitude questionnaire-c Diabetes assertiveness Test-c	children with diabetes had less fear of failure than normal controls children with good metabolic control more fear of failure, fewer problems on diabetes assertiveness test and less diabetes-related self-defense
Auslander, 1990	newly diagnosed	53	2-18	General Information and Problem-Solving questionnaires-p and c Family Inventory of Life Events and Changes-p Family Inventory of Resources of Management-p	mothers, fathers and children scored significantly higher on general knowledge test than on the problem-solving test parents of youth ages 15-18 scored lower on knowledge test than parents of younger youth age, sex, family stress, family communication abilities and financial resources are predictors of general information and problem solving

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
Hagen, 1990	diabetes non-diabetes	30 30	8-17	WISC-R-c Perceived Competence for Children-c Peabody Individual Achievement Test-c information processing tasks-c	Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report children with diabetes scored within normal range on intelligence and academic performance but evidenced some school difficulties as reflected in subscale performance as in their need of remedial education services evidence was found to suggest deficiencies in use of strategies to organize and recall information particularly those with early onset diabetes perceived self-competence and measures of family environment were notable similar across groups, families with diabetic children were more organized parents of children whose illness began prior to 5 years reported their children to have poor attention span and difficulty completing tasks
Rovet, 1990	diabetes at onset T0 1 year postdiagn T1 siblings T0	63 40	7.32 8.17 (<12)	age appropriate tasks, different for each age group (<12)	no differences between DM and S at T0 nor any specific impairment in DM predating illness diabetes mellitus no acquired impairment at T1 children early onset DM (<5 years) lower in spatial ability at T0 and T1, than children with later onset DM later onset minor decline in verbal ability asymptomatic and mild hypoglycemia correlated positively with improved outcomes in verbal area no adverse effects of severe hypo ketonuria and hospitalizations associated with lower performance IQ's 1 year after onset as was diabetic ketoacidose at onset
Auslander, 1990	diabetes	42	10	Family Environment Scale-p Piers-Harris Self Concept Scale compliance-c adjustment two questions-p and c HbA1	children from black and single-parent families were at higher risk for poorer disease control, which persisted after 2 and 3 years after diagnosis significant differences in adherence in white mothers reporting higher level of adherence
Kovacs, 1990	newly diagnosed	95	8-13	Children's Depression Inventory-c revised Children's Manifest Anxiety Scale (RCMAS)-c Coopersmith's Self-Inventory (SEI)-c Issues in coping with diabetes	illness duration and initial level of depression were predictors of later depressive symptomatology whereas little depression become somewhat more symptomatic over time levels of anxiety increased in girls, in boys decreased self-esteem remained fairly stable children perceived themselves as well-adjusted in anxiety, depression and self-esteem average levels of symptoms lower than for normative samples and lower than considered to be

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
				mellitus-child version-c	Only first authors are mentioned. c=child report, p=parent report, t=teacher, ph=physician report
Jacobson, 1990	4 year follow up diabetes	61	9-12 13-16	Nowicki & Strickland Locus of Control measure-c clinical interviews-c adherence-ph	clinically significant even with negative medical events there was generally positive self-esteem with increasing duration, implications were more upsetting and self-management tasks harder to do initial adjustment predicts later adjustment initial assessment of coping and adjustment were predictive of adherence over the 4 years psychosocial variables predicted adherence independent of age at entry preadolescents were more adherent than adolescents when diagnosed age adjustment and ego defense level accounted for 47% of the variance in adherence
Hauser, 1990	4 year follow up diabetes	52	9-16	Family Environment Scale (FES)-p/c health providers ratings of adherence-ph	family support was associated with short-term and long-term adherence family conflict adversely influences adherence parents' ratings of family organization in first year associated with higher levels of adherence youngsters perception of organization not linked with short-term or long-term adherence no relation between family valuing of independence and adherence no relation between perceived family control and adherence few connections between change in adherence and early family environment
Reich, 1990	diabetes non-diabetes	43 14	6-14	blood glucose Halstead-Reitan Neuropsychological Test battery for Children-c Wechsler-c Klove-Matthews Motor Steadiness Battery-c	mild hypo involves reduced motor performance attention and memory even after physical symptoms have subsided significant decrease in performance on neuro psychological tests after hypo few differences in overall function effect of episode centered on immediate effects rather than long-term effects
Puczynski, 1990	diabetes after hypo diabetes after euglycemia	14 10	7-15	Blood glucose Halstead-Reitan Neuropsychological Test Battery for Children-c Wechsler-c Klove-Matthews Motor Steadiness Battery-c	significant differences in five of 12 tasks, most apparent on memory and concentration no differences were noted when second trial scores of both groups were compared when they were tested after euglycemia a discrepancy may exist between the resolution of physical symptoms and restoration of cognitive function after mild hypoglycemic episodes

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
Band, 1990	diabetes preformal group diabetes formal operational group	32 32	8.8 14.6	interview-c Medical Adjustment Scale-physician Socio-Behavioral Adjustment Scale-p Connors Parent Questionnaire-p	Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report Groups differ in coping approaches, factual knowledge and medical adjustment as rated by the staff formal group: knowledge was related to medical adjustment formal group:primary-secondary coping style; perceived coping efficacy, diabetes knowledge each significantly associated with diabetes preformal group: perceived control was related to adjustment all child characteristics related to adjustment measures no relationship between a child variable and an adjustment measure replicated across the two cognitive levels
Grey, 1991	diabetes mellitus	103	8-18	Child Adolescent Adjustment Profile (CAAP)-c Self Perception Profile-c State Trait Anxiety Inventory for children-c Child Depression Inventory-c Coping Orientation for Problem Experiences-c Coping Health Inventory for Parents-p Self-Care Questionnaire-c Life Events Checklist-c	preadolescents were less depressed, less anxious, coped in more positive ways, had fewer adjustment problems and were in better metabolic control than adolescents metabolic control worsen with increasing age older children more avoidance coping children who cope avoiding and were more depressed were most likely to have problems in adjusting and metabolic control investing in close friends related to poor metabolic control parental coping styles no relationship to children's coping or adaptation
Kovacs, 1992	diabetes mellitus	95	8-14	interview-p and c Diabetic Management Information Sheet Interview Schedule for Children-c Social Competence Scale of Child Behavior Checklist-p Coopersmith Self-Esteem Inventory-c WISC-R vocabulary test-c Family Concept Inventory	29.5% serious noncompliance with medical regimen noncompliance typically started at 15 years old, 3.5 years after onset late adolescent most amount of time being non-compliant social competence, self-esteem, aspects of family functioning, initial psychiatric status did not predict noncompliance noncompliance was associated with psychiatric disorder later

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures
Weist, 1993	diabetes mellitus	56	8-19	Family Environment Scale (FES)-p Diabetes Family Behavior Checklist (DFBC)-c and p Child Behavior Checklist (CBCL)-p State Trait Anxiety Inventory for children (STAI)-c and p Multi dimensional Health Locus of Control Scale (MHLC)-c KIDCOPE-c Diabetes Knowledge Test (DKT)-c and p Diabetes Care Profile (for adults)-c (p assisted) Structured interviews with youths
Safyer, 1993	diabetes: preadolescents adolescents	42 (22) (20)	9-17	Family Environment Scale-c and p Diabetes Adjustment Scale-c
Auslander, 1993	diabetes	53	2-18	Family Inventory of Life Events and Changes-p Family Inventory of Resources for Management-p Coping Health Inventory-p diseases-related knowledge-p and c
Reid, 1994	children-diabetes	27	8-18	Test of Diabetes Knowledge-c Self-Report Coping Measure-c

Only first authors are mentioned.
c=child report, p= parent report, t= teacher, ph=physician report

Results:

younger children were more adherent
older children more knowledge, nonoptimal control higher knowledge
older children had more controlling family environments
more optimally controlled children had more controlling families
no differences between optimally and nonoptimally controlled youths on measures of anxiety, child behavior and coping style
optimally controlled youths higher on powerful others scale

successful initial diabetes adaptation depended upon family environment that is highly cohesive and organized
diabetes adjustment more strongly associated with family cohesion for pre-adolescent youngsters in contrast to adolescents
family organization is important for both pre-adolescents and adolescents

higher levels of family stress and lower levels of family resources as reported by fathers associated with poor metabolic control
family stress and resources more strongly correlated to metabolic control than disease parameters

Knowledge of diabetes not significantly related to adherence when controlling for age and duration

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
	adolescents- diabetes	29		Metabolic control (HbA1c) Diabetes Management Information Sheet-p Diabetes Regimen Responsibility Scale-p	Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report Results: higher levels of avoidance-coping strategies related to poorer metabolic control and poorer adherence approach coping predicted diet adherence, not fingerpricks coping predicted child responsibility for fingerpricks higher levels of avoidance higher levels of responsibility for diet
Kovacs, 1994	8 year follow up	57	8-14	Four-word short term memory test-a Logical memory test-a Vocabulary test of WISC-R-c Vocabulary test of WAIS-a Depression Anxiety Score-c	after 8 years of having diabetes mellitus, youths performed within average range on a test of verbal memory level of verbal association was lower than expected, short term memory was mildly impaired
Grey, 1995	diabetes without diabetes 6 weeks after diagnosis, 3/6/12/24 months	89 53	8-14	Children's Depression Inventory-c State Trait-Anxiety Inventory-c general health scale (Rand)-c Child and Adolescent Adjustment Profile-c Self Perception Profile-c	initially children with diabetes mellitus more depressed, more dependent, more withdrawn 1 year post-diagnosis, no significant differences in psychosocial status 2 years, depression, dependency and withdrawal significantly higher than in peers without diabetes self-perceived competence remained stable in both groups
Burleson Davis, 1995	diabetes mellitus	80	10-14	Diabetic control (HbA1c) Child Behavior Checklist p Diabetic Adaptation Scale (DAS) c Coppersmith Self Esteem Inventory c Revised Children's Manifest Anxiety Scale (RCMAS) c Demographic, medical, diabetic adherence and parents attitudes p	a slight trend on the CBCL toward increased psychopathology relative to normative population higher self esteem than normative population anxiety lower than the normative population family size and duration influence on diabetic control, larger family size better control, longer duration worse control competence/adherence influence on diabetic control weaker correlation between diabetic adherence and metabolic control

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures	Results:
Reid, 1995	diabetes mellitus	56	8-18	Self-report Coping-c Interview-c Grade Point Average-p Child Depression Inventory-c or Reynolds Adolescent Depression Inventory Social Skills Rating System SSRS-t Self-Rated Coping Effectiveness-c	Only first authors are mentioned. c=child report; p= parent report, t= teacher, ph=physician report children used approach-coping strategies more often than adolescents higher levels of approach coping and lower levels of avoidance coping were generally related to more positive outcomes, adjustment coping strategies were within normal range
Simell, 1995	two years follow up short term treatment at diagnosis (25m, 20f) long term treatment (27 m 27 f)	54	0-15	interview with family living with diabetes questionnaire (this study)-m+f	more than two-thirds of the families had diabetes mellitus related fears over half of both groups confident about care after 1 year children in long-term doing better at school none of the families expressed that diabetes mellitus caused decreasing child's success at school three families had minimum scores in well-being of which team was unaware (of two families they were aware) fathers scored higher than mothers except in adherence to care glycemic control and families well-being not being depended on father's involvement families more hobbies than earlier
Northam, 1996	diabetes	106	1-14	Child Behavior Checklist-p General Health Questionnaire GHQ-28-p Family Adaptability and Cohesion Evaluation Scales FACES-III-p	children <4 did not differ from CBCL norms at any time children between 4-11 above norm at M1, M2 significant below onset of diabetes for schooled children associated with mildly elevated levels of mother-reported psychosocial distress children some distress at time of diagnosis, however largely resolved at M2 family function respondents differ from distress, respondent, child-age and SES anxiety reported by father and mother, social disruption for some groups complex findings related to family function
Hanson, 1996	diabetes m1 m2 m3	228 270 256	4-20	glycemic control self-care adherence interview dietary recalls and logs physical activities recalls& logs	reliable and significant relationships between glycemic control, SCAIC, overall quality of youth's dietary intake consistency in dietary patterns most strongly associated with glycemic control

Table 2.2: Clinical studies on the functioning of children with diabetes mellitus

Authors	Group	N	Age	Measures
Sterling, 1996	diabetes healthy	54 54	6-12	Child Behavior Checklist-p Comprehension subtest of WISC-R-c
Rovet, 1997	diabetes healthy	103 100	9.3- 18.3	

Only first authors are mentioned.
 c=child report p= parent report, t= teacher, ph=physician report

Results:
 children with diabetes mellitus utilized different types of qualitative statements than healthy peers, revealing a more sophisticated empathic responding, less self-focused statements of personal distress
 groups didn't differ on empathy continuum
 children with diabetes higher scores on behavior problems

diabetes subjects differed from control subjects in the select component of attention, poorer performance of children with early onset (<6 years)
 those having had hypo lower verbal IQ and greater difficulty with select, focus and inhibit attentional components whereas sustain suppress and shift attentional focus were unaffected
 higher blood glucose levels associated with less adequate ability to inhibit impulsive responses ;
 onset age and concurrent blood glucose predicted inhibit and focus

Table 2.3: Clinical studies among children with epilepsy				
Authors	Group	N	Age	Measures
Dorenbaum, 1985	epilepsy	38	6-16	Child Behavior Checklist-p
Corbett, 1985	complicated epilepsy	312	5-18	Rutter's scale-p and t EEG physical examination
Rodin, 1986	epilepsy	64	5-16	Wechsler reassessments after at least 5 years-c
Noda, 1989	epilepsy	19	10-13	WISC-R-c Benton Visual Retention Test-c Wechsler Memory Scale-c
Blenmow, 1990	epilepsy healthy	69 69	7-18	cognitive test battery (Meer & Bosch)-c
Hoare, 1991	epilepsy	108	5-15	semistructured interview-p Edinburgh Parental Attitude Scale to Epilepsy (EPASE)-p Rutter Parent and Teacher Questionnaire-p Piers-Harris Questionnaire-p Self-Administered Questionnaire (SADQ)-p GHQ-30-p
<p>Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report</p> <p>Results the highest risk of maladjustment was within social functioning, as children reach adolescence most problems related to school as a group did not differ from normgroup, but evidence for behavioral maladjustment in individual children</p> <p>significant association between long-term anticonvulsant therapy and cognitive deterioration association between specific behavioral problems and focal cerebral dysfunction no association between medication and overall behavioral deviance</p> <p>in general seizures states had been improved, 50% in remission for between 2 and 8 years verbal and performance areas could be affected differentially, gain in one could be offset by a loss in the other those whose seizures remained statistically significant decrease in performance IQ whereas it was stable or increased for patients in remission decreased IQ indicated slower mental growth rather than loss phenobarbital inversely correlated with IQ</p> <p>early onset of epilepsy, regardless type of epilepsy, places child at risk of cognitive dysfunction polytherapy as well as more than a 3-year duration of epilepsy , impair visual short-term memory of cognitive functions but not the auditory short-term memory or IQ</p> <p>interim analysis of results suggests that short-term memory is decreased in all sub-groups of children being treated for epilepsy</p> <p>high rates of disturbance (50%) on both the Rutter Parents' and teachers' scales children were regarded by their parents and by other adults as predominantly worried, fearful, miserable and solitary, rather than as acting out or destructive similar rates of disturbance in boys and girls total score and subscale scores on the Piers-Harris measure of self-esteem were not significantly different from values of the general population SADQ indicates that children with epilepsy differ from general population on affection subscale for boys secondary schoolage girls, communication subscale for secondary school-age boys and assistance subscale for primary school-age girls, families of children in special schooling more</p>				

Table 2.3: Clinical studies among children with epilepsy				
Authors	Group	N	Age	Measures
				<p>Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report</p> <p>Results stress than families of children attending normal schools more mothers with primary school-age children GHQ scores above cut-off point compared to mothers of secondary school age children. parents had no significant more psychiatric morbidity than adults in normal population demographic and family variables, epileptic variables, maternal attitude to epilepsy and individual characteristics of the child are associated with poor psychosocial adjustment early onset and high fit frequency adversely affected the likelihood of disturbance maternal attitudes towards epilepsy were also associated with an increased risk of disturbance cognitive and education attainments not associated with psychosocial morbidity</p>
Suurmeyer, 1991	epilepsy healthy (5 year follow up)	136 109	11 (M1)	<p>oral interviews-many subjects- p 5 years later: mailed questionnaires on medical situation and educational situation</p>
Austin, 1992	epilepsy	127	8-12	<p>five variables predicted behavior problems: female gender, family stress, family master, extended family social support, seizure frequency children who were experiencing behavior problems tended to have poorer seizure control, to be in troubled families in which mothers were receiving less support than needed from relatives. family variables are important correlates of behavior problems</p>
Huberty, 1992a	epilepsy	136	8-12	<p>no significant relationships between seizure variables and achievement scores nearly 40% had been retained at least once before sixth grade children with secondary generalized seizures were more likely to have been retained in grade but no relation between being retained and seizure type</p>
Huberty, 1992b	epilepsy	131	8-12	<p>age at onset was not related to achievement or adaptive skills seizure frequency seems to be related to academic and affective problems academic function within normal range seizure disorder associated with a diffuse or multifunctional brain insult can produce problems in achievement and school adaptation</p>

Table 2.3: Clinical studies among children with epilepsy					
Authors	Group	N	Age	Measures	Results
Hoare, 1993	epilepsy	108	5-15	modified impact of epilepsy schedule-p Holroyd questionnaire on Resources and Stress-p	Only first authors are mentioned. c=child report p=parent report, t= teacher, ph=physician report epilepsy greatest impact on children who had intractable epilepsy and additional disabilities early onset epilepsy accompanied by additional disabilities had a widespread adverse effect on child's and family's quality of life and overall adjustment adverse impact was clear on management of epilepsy (side effects), adjustment, development and restrictions in family life and activities
Jambaque, 1993	epilepsy healthy	60 60	7.5- 14.5	Wechsler-R-c Signoret's Memory Battery-c	epilepsy may contribute to memory deficits in children no evidence of memory deficits in daily life but difficulties in academic achievement that reflected their memory impairment anticonvulsant drugs, seizure frequency and age at onset not individually influence on memory type of epilepsy played major role in pattern of memory impairment: idiopathic epilepsy slight depression of visual memory partial epilepsy more severe memory disorder left and right temporal lobe epilepsy marked memory deficits related to hemispheric specialization
Aldenkamp 1993	epilepsy healthy	83 83	7-18	Finger Tapping Test-c Simple Reaction time Measurement-c Binary Choice Reaction Test-c Computerized Visual Searching Task-c	results showed significant improvement attributable to drug withdrawal on only one of the cognitive tests (psychomotor speed), suggesting impact of ASED treatment on higher order cognitive function is limited patients with a former diagnosis of absence seizures show lower scores both at baseline and after drug withdrawal before and after drug withdrawal differences between epilepsy and healthy on information processing speed and recognition of words before differences between epilepsy and healthy for tapping rate with dominant hand
Aldenkamp 1994	epilepsy healthy	83 83		Holmifrid Quality of Life Inventory-ph read out the questions for children and parents separately	after all treatment had been stopped and children were cured, parents of epileptic children still report epilepsy-related problems, remained worried after treatment, reported new complaints such as increased aggressiveness epileptic youths showed abrupt adaptation to new situation, a superpositive quality of life and a cessation of complaints
Mitchell, 1994	epilepsy	157	4-13	McCarthy Scale of Children's abilities, WISC-R-c CHLA Behavior Questionnaire-	seizure history was best predictor of seizure control and general health status severity and controllability remained stable over the observation period sociocultural variables were predictors of parental anxiety and negative attitudes

Table 2.3: Clinical studies among children with epilepsy				
Authors	Group	N	Age	Measures
				<p>P Coddington Life Events Scale for Children-c Child Behavior Checklist-p Families attitudes-p</p>
Schmidt, 1995	epilepsy	19	6-14	<p>General Questionnaire for this study-p Labyrinth Lerntest nach Milner (1965)-p Continuous Performance Test (CPT)-c Reaction test-c Rhythm tapping-c EEG</p>
Carlton-Ford, 1995	part of large NHIS study: active epilepsy inactive epilepsy no history of epilepsy	116 0	6-17	Interview-p
Williams, 1995	epilepsy head injury substance abuse psychiatric group	43 30 61 19	5-17	Wide Range assessment of Memory and Learning (WRAML)-c
<p>Only first authors are mentioned. c=child report p= parent report, t= teacher, ph=physician report</p>				
<p>Results variation in medical or attitudinal outcomes not influenced by IQ or behavioral problems</p>				
<p>according to subjective parent reports there is a correlation between Valproat Therapy and development of stronger attention and concentration disturbances</p>				
<p>children with history of epilepsy have lost more days to illness previous year adults perceive children with history of epilepsy as more likely to have a life-threatening illness and in poorer general health and more clumsy children with active epilepsy and children with inactive epilepsy fare about equally children with any history of epilepsy fare worse than children without any history of epilepsy: they have significantly higher odds of high levels of home behavior problems, depressed mood and impulsiveness, the odds of being the most impulsive are over five times as high for children with a history of epilepsy a history of epilepsy was not significantly related to school behavior problems demographic and family variables moderate only a small part of epilepsy's effect combination of family process and occurring conditions appears to produce epilepsy's apparent effect findings suggest that poor social and psychological adjustment probably results from self-fulfilling prophecy children with epilepsy had visual memory skills within the low average range, whereas the remising groups had those skills in the average range all children and children with epilepsy most had difficulty on the letter/number memory test which has been postulated as a measure of auditory attention skills children with epilepsy more variability on both verbal and visual memory tasks-when repetition and drill were given they had better scores-> increased problems with both attention and memory</p>				

Table 2.4: Clinical studies on the functioning of children with juvenile chronic arthritis				
Authors	Groups	N	Age	Measures
Billings, 1987	rheumatic healthy	95 93	1-18	Health and Daily Living Form-p Health Assessment Questionnaire-c and p
Thompson, 1987	juvenile chronic arthritis	23	4-16	Varni/Thompson Pediatric Pain Questionnaire-c and p and ph Family Environment Scale-p Child Behavior Checklist-p disease activity-ph
Daniels, 1987	juvenile chronic arthritis siblings healthy	93 72 93		Health and Daily Living Form-p Child Health Assessment Questionnaire-p Family Environment Scale-p Family Effects of Illness questionnaire-p
Varni, 1988	juvenile chronic arthritis	23	4-16	Varni/Thompson Pediatric Pain Questionnaire-p and c Family Environment scale p-p Family Relationship Index Child Behavior Checklist-p Child Activities of Daily Living Index-p Disease Activity Index ph Family Information Form p
<p>Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report Results: Physical Cognitive Social Emotional subjects in severe group more parent-reported psychological and physical problems than mild patients and healthy controls mothers and fathers relatively high agreement in reporting severe group missed more school due too illness than mild group older severe children more likely to miss school and participate in fewer activities than controls older children report comparable mood and functioning in other areas parents reported more physical problems and mood-related disturbance than their children did whereas there is reasonable agreement in child-parent report</p> <p>strong association between parent, child and physician ratings on pain children as young as 5 reliable judges of pain intensity pain ratings low, although higher pain scores last week raised somatic complaints on CBCL and fluctuations on social competence of CBCL cohesion and expressiveness higher and conflict lower in these families family environment and child psychological factors were found to interact with specific disease parameters to modulate pain experience</p> <p>higher parental depression and medical symptoms and more family stressors sibling problems and burden of illness on family predicted more problems risk and resistance similar for healthy siblings, although cohesion and expressiveness were more important and illness burden less salient mothers' depression lack of family cohesion, expressiveness related to more adjustment problems among healthy children</p> <p>child psychological adjustment, family psychosocial environment, pain in combination with disease activity statistically predict functional status model most powerful for predicting children's ADL</p>				

Table 2.4: *Clinical studies on the functioning of children with juvenile chronic arthritis*

Authors	Groups	N	Age	Measures
Ungerer, 1988	juvenile chronic arthritis primary school high school young adult	363 (109) (163) (91)	7-31 7-13 12- 20 16- 31	for age-categories other instruments-c Piers-Harris Self-concept Scale-c Self-Description Questionnaire-c severity and general questionnaires-p
Konkol, 1989	arthritis	50	2-22	7 open ended questions-c and p and siblings
Vandvik, 1990	juvenile rheumatic	106	1-17	interview-p and c Child Assessment Schedule-c Children's Global Assessment Scale-based on interviews Child Behavior Checklist-p
Ennett, 1991	juvenile chronic arthritis	38	7-13	Disease severity-ph disease experience by items constructed for the study -c Self Perception Profile for Children-c and p

Only first authors are mentioned.

c=child report p= parent report, t= teacher report, ph=physician report.

Results: Physical Cognitive Social Emotional

psychologic functioning and disease severity were related to adjustment in primary school and high school
measures of social relations related to adjustment in high school
relations among psychological functioning, social relationships and adjustment in young adults were minimal
levels of disease severity associated with financial concerns, emotional problems and physical strain in parents of high school and young adults

diverging views of family members were found regarding perceptions and impact of juvenile chronic arthritis
siblings and children but not parents mentioned physical problems
children are concerned about school, siblings about understanding emotions, parents about personal stress and stresses on relationships
parents wanted more knowledge, children not children and siblings are scared for death

half of the patients received psychiatric diagnosis, most of affective or anxiety types, while behavior disorders were underrepresented
psychosocial dysfunction of at least mild severity was found in 64%
polyarthritic group negative correlation between CGAS and severity of disease
total behavior scores close to American and Dutch norms

gender not significantly associated with variables
age was related to emotional functioning and social acceptance
modest correlation mother-child on perceived competence, and perceptions of experience:
mothers rated children's competence more negatively than the children rated themselves;
children reported day-to-day experiences significant worse than mothers did whereas mothers described effects on family as worse
no significant differences were found between children and mothers in perceptions of emotional or social impact
disease severity related to athletic competence and perceptions of their physical attractiveness
disease experience measures also correlated with perceived competence and social acceptance
mean scores on self perception less than reference group

Table 2.4: *Clinical studies on the functioning of children with juvenile chronic arthritis*

Authors	Groups	N	Age	Measures
Harris, 1991	arthritis healthy	12 12	6-11	Perceived competence(Harter)-c Coping scale-p Family crisis oriented of personal evaluation Scale-p Family environment Scale Child and Adolescent Coping Inventory Child Behavior checklist-p Family effects of illness Scale-p Faschingbauer abbreviated MMPI (FAM)-p Observations of types of peer interactions frequency scoring
Timko, 1992	arthritis	147	M= 11.1	Health and Daily Living form-p Family Effects of illness Scale-p Family Environment Scale-p Child Health Assessment Questionnaire-p+t-c Functional Disability Index-p
Daltroy, 1992	arthritis	102	4-16	CBCL-p Profile of Mood States-p
Berry, 1993	juvenile chronic arthritis	54	6-17	items on juvenile chronic arthritis scoring on Bibace and Walsh-c

Only first authors are mentioned.

c=child report, p= parent report, t= teacher report, ph=physician report

Results: Physical Cognitive Social Emotional

juvenile rheumatic disease (JRD) is not associated with detrimental psychosocial outcomes
children with rheumatic disease and parents displayed levels of competence, adjustment, and
stress comparable to those of healthy controls
children's peer relations are very similar to that of healthy controls
JRD children and families actively utilize multiple coping strategies

risk, resistance and disease-related factors stable and predicted psychosocial outcomes
prior risk and resistance factors are less successful predictors than concurrent risk and
resistance factors, however mother's depressed mood was stable
mothers who were not depressed and had a sense of mastery over their child's illness
appeared to aid adjustment

on average, parental stress was lower than reference norms
POMS distress was correlated with behavior problems but not with children's social
competence.

older age was associated with more behavior problems in males
disease severity and disease activity were also associated with behavioral problems
no single predictor variable especially strong in predicting social competence
older age and shorter duration, associated with poorer social competence
teenagers especially those with recent onset and mild activity may be at increased risk
modest excess of behavioral and social problems, parents however not unduly distressed

children's understanding of illness followed a developmental progression, with older children
having a more sophisticated understanding
conceptual level was a better predictor than age
significant number of children functioning below age-level
majority understanding at the concrete operational level of cognitive development

Table 2.4: Clinical studies on the functioning of children with juvenile chronic arthritis			
Authors	Groups	N	Age
Doherty, 1993	juvenile chronic arthritis	20	8-16
			Measures Child Health Assessment Questionnaire-c and p
Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report Results: Physical Cognitive Social Emotional a high level of agreement between children and their mothers with respect to disability no correlation between children and mothers on pain mother's ratings of pain and disability highly correlated whereas children's was not 6 children negative attitude towards juvenile chronic arthritis possibly association with disagreement between parents and children			
Timko, 1993	study 1 study 2 4 years later	204 172	9.3
			Health and Daily Living Form-p Depressed Mood-p Drinking problems-p Family Effects of Illness Scale (FEI) Social Activities with friends-p Close relationships-p Child HAQ-p or c> 10 yr. Functional disability Pain CBCL-p
older patients experienced more pain patient's functioning at time 1 was a strong predictor of functioning 4 years later-in the absence of intervention mother's depression related to having patients more problems more social activities mother, more social competence among patients father's initial depression predicted patients having more functional disability and father's personal strain predicted patients having less social competence father's drinking problems were related to poorer disease-related outcomes, but also to more social competence			
Ross, 1993	juvenile chronic arthritis	56	7-17
			Children's reported pain c 28 day diary Disease characteristics-ph Spielberger Trait/State Anxiety Scale-c-interview Child Depression Inventory-c-interview CBCL-p Lanyon Psychological Screening Inventory (adult scale for p) Family Environment Scale-p
child's distress, maternal distress and family harmony/conflict had moderately high statistically sign partial correlations with child's reported pain. substantial contribution of psychological variables to reported pain greater family harmony was related to higher reported pain ->behavioral treatment programs for pain needed			
Balldam, 1995	juvenile chronic arthritis	29	7-16
			Juvenile Arthritis Functional Assessment Report-c and p pain and stiffness VAS-c Steinbocker Functional Class-c,p, researcher
JAFAR score correlated with stiffness, pain score, Steinbrockers classification psychological dysfunction in 8 children (27%) only one child high score on Birlson scale to suggest depression self concept was scored as healthy population no correlation between psychological scores and functional measurements			

Table 2.4: Clinical studies on the functioning of children with juvenile chronic arthritis		Only first authors are mentioned. c=child report p= parent report, t= teacher report, ph=physician report		
Authors	Groups	N	Age	
			Measures	
Hagglund, 1995	arthritis	60	7-17	Rutter Parental Screening Questionnaire A-p Rutter B-teacher Birleson depression invent.-c Lippstitt self-concept scale-c Hopelessness Scale for children-c Sadness scale from the Differential Emotions Scale IV-c Social Support Questionnaire revised-c Pain visual analog Scale-c
Mallison, 1996	arthritis healthy	31 16	8-17	physical tests-c pain-scale from the Pediatric Pain Questionnaire-c Juvenile Arthritis Functional Assessment Report-c and p Self Perception Profile for Children Questionnaire-c
Results: Physical Cognitive Social Emotional				
Rutter A scores were high in 21% in comparison with 13% in controls parents were accurate in reporting level of physical functioning as measured by child and parent report				
demographic variables, disease status, social psychological measures did not predict pain pain correlated with disease duration and age; report more pain as they grow but less pain the longer they have JCA				
no significant differences between patients and controls for either aerobic or anaerobic fitness negative association between peak O ₂ uptake and sum of skinfolds only a small minority rated athletic competence lower than norm and all but one rated global self-esteem at or above normative mean data findings suggest that children with arthritis are more realistic in their appraisal of competence than healthy peers				

Table 3.1: Health status and quality of life instruments for children – an overview

Name of instrument	Group	Age	Item n	R	Dimensions	Ca	Remarks
[1] HIS Measure Eisen, 1979	healthy	0-13	23	P	mental health (12 items), social health, general health ratings (7 items) 2 scores: mental health, general health ^{1,3,4}	g	two versions: 0-4 5-13 1 month, 3 months, and in general
[2] RAND Lewis, 1989	healthy chronically ill	0-13	4	P	general health and distress caused by health problems, one score ¹	g	3 months and in general validated in the Netherlands (Post, 1998)
[3] Functional Status Measure FS-IIR Stein, 1990	healthy chronically ill	0-16	14	P	one health dimension, impact of illness on child's physical, social and psychological functioning ^{1,3,4}	g	full 43-item version available last 2 weeks validated in the Netherlands (Post, 1998)
[4] Health Resources Inventory Gesten, 1976	healthy	1-3 gr	46	t	good student, gutsy, peer sociability, rules, frustration tolerance, total score ^{2,3,4}	g	primary grade teacher measure 1-3 graders at primary school
[5] Child Health Questionnaire Landgraf, 1996	healthy chronically ill	5 and older	98	P ch	limitations in physical activity, limitations in school or social activities, limitations in family activities, bodily pain, general mental health, general behavior, satisfaction with self, family, friends and life in general, general health perceptions. ^{1,2,3,4}	g	parent form includes 98 items, the child form 87. A Dutch translation is available (Raai, GCID-Rotterdam).
[6] Generic Child Quality of Life Measure (GCQ) Collier, 1996	healthy chronically ill	6-16	22	ch	child is first asked to respond to all questions relating to the child they felt is most like themselves, then the children answer all the questions they would most like to be one score, provided by the total sum of discrepancies between the two dimensions ^{3,4}	g	perceived self-score story format to start with
[7] TACQOL Verrips, 1998	healthy chronically ill	5-15	56	P ch	physical complaints, motor functioning, autonomous living, social functioning, cognitive functioning, positive moods, negative moods, profile scores ^{1,2,3,4}	g	frequency and evaluation for each relevant item

Table 3.1: Health status and quality of life instruments for children – an overview

Name of instrument	Group	Age	Item #	R	Dimensions	Ca	Remarks
[8] Generic Self assessment of HRQOL (16D) Apajasalo, 1996	healthy chronically ill	12-15	16	ch	profiles and summary score, 16 mc items each representing one related health dimension, f.i. mobility or friends ^{1,2,3,4}	g	based on adult 16 D measure
[9] Child Quality of Life Questionnaire (CQOL) Graham, 1997	healthy psychiatric disorder chronic physical disorder mental retardation	9-15	45	p ch	fifteen areas of daily life: getting about and using hands, doing things for self, soiling or wetting, school, out of school activities, friends, family relationships, discomfort, worries, depression, seeing, communication, eating, sleep, appearance ^{1,2,3,4}	g	three items each domain: how well, satisfaction and upset reaction 1 month, 10-15 minutes 7-point scale test-retest reliability low
[10] RAHC Measure of Function (MOF) Dossetor, 1996	pediatric patients	10-	1	ph p	generic measure, 10 categories, one level of dependency in health score related to functioning, tick only one score (grading severity) ^{1,2,3,4}	g	two versions: Clinical Rating Scale -ph Family Rating Scale-p modified from Child Global Assessment scale, a psychiatric instrument for children
[11] Pediatric Symptom Checklist Jellinek, 1988	pediatric patients	6-12	35	p	age-appropriate goals in school, play, peer relationship and family life, mood total score and cut-off score ^{1,2,3,4}	g	
[12] Paediatric Asthma Quality of Life Questionnaire (PAQLQ) Juniper, 1996a	asthma	7-17	23	ch	activity limitations, symptoms, emotional function and total score of items that children have identified as troublesome total score and profile ^{1,3,4}	d	in the activity domain, three items are individualised
[13] Childhood Asthma Questionnaire (CAQ) French, 1994	asthma	4-16	48	ch	generic as well as disease specific, active quality of living, passive quality of living, distress, severity ^{1,3,4}	d	frequency and evaluation (smiley faces scale) separate versions for distinct age categories

Table 3.1: Health status and quality of life instruments for children – an overview

Name of instrument	Group	Age	Item n	R	Dimensions	Ca	Remarks
[14] Asthma Symptom Checklist (CASL) Fritz, 1989	asthma	6-18	47	ch p	general physical symptoms, panic/fear, hyperventilation/irritability ^{1,4}	d	modified from adult version parents were asked to rate items as they thought their children experienced them; however factors not the same content CASCL greatest potential for use as a parent observation form
[15] A life Activities Questionnaire for Childhood Asthma Creer, 1993	asthma	5-17	71	ch	physical activities (20 items), work activities (4 items) outdoor activities (16), emotions and emotional behavior (6 items), home care (11 items), eating and drinking (5 items) miscellaneous (9 items), one score possible ^{1,4}	d	parents could read the items and instructions for their child adapted from the adult-version (Life Activities for Adult Asthma)
[16] Asthma Problem Behavior Checklist (APBC) Creer, 1983	asthma	5-15	27	p or ch	general information, identification of behaviors important in prevention of attacks, attack-related behavior, behavioral consequences, socio-economic factors-behavioral profile ^{1,4}	d	
[17] Modified Quality of Life Measure for Youths Ingersoll, 1991	diabetes mellitus	10-20	52	ch	diabetes impact, disease-related worries, diabetes life satisfaction total score possible ^{1,2,3,4}	d	developed from adult questionnaire
[18] Quality of life scale Wildrick, 1996	epilepsy	8-18	25	ch	self concept (5 items), home life (5 items), school life (9 items), social activities (4 items), medication issues (2 items) ^{1,2,3,4}	d	adapted from the Quality of Life in Epilepsy (QOLIE-89)
[19] Making Waves Survey Brown, 1994	epilepsy	0-18	30	p	seizure variables, medication, attitudes towards seizures, medication, communication with doctors, perceived effect of epilepsy on activities, relationships, school life and personal self-esteem ^{1,2,3,4}	d	

Table 3.1: Health status and quality of life instruments for children – an overview

Name of instrument	Group	Age	Item	R	Dimensions	Ca	Remarks
[20] Impact of Childhood Illness Scale Hoare, 1995	epilepsy	6-17	30	p	impact of epilepsy and its treatment (5 items), impact on child's development and adjustment (10 items), impact on parents (5 items) impact on family (10 items) ^{1,2,3,4}	g	each item not only frequency, also degree of importance (concern) two forms:boys and girls
[21] Childhood Health Assessment Questionnaire (HAQ) Doherty, 1993; Singh, 1994	juvenile chronic arthritis	1-19	35	p ch	dress and grooming, arising, eating, walking, hygiene, reach, grip, activities-two scales: disability (8 functional areas) and discomfort (1 item) ¹	d	less than 10 minutes to complete parallel parent and child questionnaire at least one item for each functional area is relevant for all ages
[22] The Juvenile Arthritis Quality of Life Questionnaire (JAQQ) Duffy, 1997	juvenile chronic arthritis	1-18	74	p ch	gross motor function (17 items), fine motor function (16 items), psychosocial function (22 items), general symptoms (19 items)no total score also a pain score as a result of the disease ^{1,2,3,4}	d	less than 20 minutes to complete; above 9 years of age also patients report asked to select 5 items of individual priority for greater responsiveness in each dimension (with which the child has most difficulty) younger than 10 years pain dimension with faces scale
[23] Juvenile Arthritis Functional Status Index (JASI) Wright, 1996	juvenile chronic arthritis	8-19	100 (I)	ch	Part I consists of 100 items divided into 5 activity categories: self-care, domestic, mobility, school extracurricular; part I is scored out of 100 points where each of five categories is calculated from a maximum of 20 points; Part II: priority function section identifies and scores (5) important areas for improvement ¹	d	mean time 50 minutes to complete; part two needs the assistance of an interviewer and requires less than 15 minutes

Table 5.6: Medical care

	Category	Asthma n(%)	IDDM n(%)	Epilepsy n(%)	JCA n(%)	
Number of visits paid to specialist/pediatrician during the past six months due to the chronic illness	0	49 (23%)	0	9 (10%)	3 (5%)	
	1	62 (29%)	18 (13%)	35 (37%)	5 (9%)	
	2	62 (29%)	75 (56%)	21 (22%)	18 (31%)	
	3	17 (8%)	22 (16%)	10 (11%)	11 (19%)	
	>=4	21 (10%)	19 (14%)	20 (21%)	21 (36%)	
	at least once	162 (77%)	134 (100%)	86 (91%)	55 (95%)	
	N	211	134	95	58	
Number of visits paid to GP during the past six months due to the chronic illness	0	130 (53%)	107 (84%)	73 (86%)	51 (88%)	
	1	49 (20%)	19 (15%)	9 (11%)	4 (7%)	
	2	23 (9%)	2 (2%)	2 (2%)	1 (2%)	
	3	14 (6%)	0	1 (1%)	1 (2%)	
	>=4	31 (13%)	0	0	1 (2%)	
	at least once	117 (47%)	21 (16%)	12 (14%)	7 (12%)	
	N	247	128	85	58	
Days spent in hospital during the past six months due to the chronic illness ¹	0	190 (94%)	103 (81%)	94 (90%)	51 (86%)	
	1	3 (1%)	3 (2%)	4 (4%)	2 (3%)	
	2	1 (0%)	1 (1%)	1 (1%)		
	3	1 (0%)	2 (2%)	2 (2%)	3 (5%)	
	4			1 (1%)		
	5	2 (1%)	1 (1%)	1 (1%)	2 (3%)	
	6-14	6 (2%)	11 (9%)	1 (1%)	1 (2%)	
	>14	1 (0%)	5 (4%)	1 (1%)	1 (2%)	
	at least once	14 (7%)	23 (18%)	11 (10%)	9 (15%)	
		N	204	127	105	59
Other care used during the past six months	No	215 (81%)	105 (76%)	83 (81%)	26 (44%)	
	Medical	31 (12%)	10 (7%)	3 (3%)	17 (29%)	
	alternative med	2 (1%)	1 (1%)	1 (1%)		
	psychologist	11 (4%)	14 (10%)	8 (8%)	2 (3%)	
	more than one	3 (1%)	3 (4%)	1 (1%)	12 (20%)	
	other	2 (1%)	5 (4%)	7 (7%)	2 (3%)	
	receiving other care	49 (19%)	33 (24%)	20 (19%)	33 (56%)	
	receiving other care due to chronic illness	31 (12%)	22 (16%)	8 (8%)	29 (49%)	
		N	264	138	103	63

¹ Results are missing from part of the sample that was provided by the EMGO Institute

Table 5.21: Correlations between parents and children in the healthy group of children and in the group of children with a chronic illness

Scales	Ill children R	Healthy children R
Prevalence		
Physical activities	.60*	.59*
Cognitive activities	.37*	.37*
Social activities	.50*	.52*
Physical complaints ¹	.61*	.51*
Happiness	.37*	.32*
Quality of performance		
Physical activities ¹	.61*	.30*
Cognitive activities ¹	.54*	.36*
Social activities ¹	.47*	.27*

¹ Spearman Rank correlations are used, on all other scales Pearson correlations were used

* p < .001

Table 5.22: Differences in mean scores between parents and children (non-paired t tests)

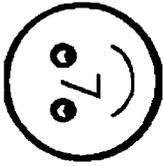
Scales		Children with a chronic illness	Healthy children
		M (SD)	M (SD)
Prevalence			
Physical activities	child report	2.63 (.55)	2.60 (.48)
	parent report	2.57 (.57)*	2.59 (.52)
Cognitive activities	child report	3.14 (.62)	3.07 (.58)
	parent report	3.04 (.58)*	3.08 (.57)
Social activities	child report	2.29 (.53)	2.37 (.48)
	parent report	2.25 (.45)	2.37 (.48)
Physical complaints ¹	child report	3.52 (.56)	3.57 (.51)
	parent report	3.58 (.53)*	3.75 (.37)*
Happiness	child report	3.11 (.63)	3.08 (.61)
	parent report	2.95 (.54)*	2.99 (.50)
Quality of performance			
Physical activities ¹	child report	3.75 (.42)	3.91 (.18)
	parent report	3.73 (.44)	3.96 (.14)*
Cognitive activities ¹	child report	3.75 (.38)	3.74 (.48)
	parent report	3.70 (.51)*	3.82 (.31)*
Social activities ¹	child report	3.85 (.33)	3.90 (.23)
	parent report	3.77 (.43)*	3.91 (.27)

* p < .01; Transformations ($X_{new} = (1/(5-X_{old}))$) were used

Note. Items were rescored so that a high score means a high quality of life, a high prevalence of activities or a low prevalence of difficulties or limitations and a low level of negative feelings

APPENDIX 2 – HAY CHILD REPORT

HOW



ARE



YOU?

April 1995, AuR-1

© University Leiden: Health Psychology
Vrije Universiteit Amsterdam:

EMGO-Institute, Dep. Of General Practice
Nursing Home Medicine and Social Medicine

HOW DO YOU ANSWER THE FOLLOWING QUESTIONS?

The questions in this book are concerned with how you are doing. Directions are given before each new section.

If you want to change your answer then please put a line through the "wrong" answer and an "x" by the right answer.

Always answer what you think. There are no correct or incorrect answers.

Please choose only one answer!

Please mark your answer by placing an "x" in the box you choose:

1. I am a Boy
 Girl

Please answer the following questions:

2. I am ___ years old
3. I am in grade ___
4. Today's date is: ___ day ___ month 19 ___

PART 1

The following questions are about how you felt during the past 7 days.

Please answer what you think and try to put an answer by each question. You can only choose one answer for each question.

Please mark your answer by placing an "x" in the box you choose:

- 1 Have you ridden your bike during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 2 Have you played outside during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 3 Have you run about during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 4 Have you joined in with gym during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often

- 5 Have you been swimming during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 6 Have you done math assignments in class during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 7 Did you manage to keep your attention on your schoolwork during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 8 Have you done language assignments in class during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 9 Have you remembered what you learned at school during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
-
- 10 Have you been to a party in the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 11 Have you played with other kids at recess during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 12 Have you played with other children after school during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 13 Have you had dinner at a friend's house during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 14 Have you visited someone, such as a family member or other children during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often

- 15 Have you felt happy during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 16 Have you felt cheerful during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 17 Have you laughed during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 18 Have you had fun during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 19 Have you felt pleased during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 20 Have you had a headache during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 21 Have you felt nauseous during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 22 Have you had a stomach ache during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 23 Did you get up feeling tired during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 24 Have you been coughing during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 25 Have you felt short of breath during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often

26 Has your breathing been wheezy during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

27 Have you woken up at night due to your asthma?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

28 Have you taken any medicine for your asthma during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

29 Did you take your medicine for your asthma with you when you went somewhere during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

30 Have you used your peak flow measure during the past 7 days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

31

Did you take your asthma medicine when other children were around during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

32 Have you worried about your asthma during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

33 Were you scared about becoming short of breath during the past 7 days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

34 Have you felt sad because of your asthma during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

35 Have you felt lonely because of your asthma during the past seven days?

- No, never
 Yes, sometimes
 Yes, often
 Yes, very often

- 36 Have you felt angry because of your asthma during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 37 Have you worried about your future due to your asthma during the past seven days?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 38 Have you felt during the past seven days that other kids did not like you as much because of your asthma?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 39 Have you felt during the past seven days that you were not able to do as much as other kids who do not have asthma?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often
- 40 Have you felt during the past seven days that you were different from other children because of your asthma?
 No, never
 Yes, sometimes
 Yes, often
 Yes, very often

PART 2

The next questions are about how well you are able to do certain things. If you don't do these things because you are not able to, you can place an "x" in the box "Not well at all".

Please mark your answer by placing an "x" in the box you choose:

- 41 How well are you able to ride your bike?
 Not well at all
 Not well
 Not so well
 Well
- 42 How well are you able to play outside?
 Not well at all
 Not well
 Not so well
 Well
- 43 How well are you able to run?
 Not well at all
 Not well
 Not so well
 Well
- 44 How well are you able to join in gym?
 Not well at all
 Not well
 Not so well
 Well

- 45 How well are you able to swim?
 Not well at all
 Not well
 Not so well
 Well
- 46 How well are you able to do math assignments in class?
 Not well at all
 Not well
 Not so well
 Well
- 47 How well are you able to keep your attention on your schoolwork?
 Not well at all
 Not well
 Not so well
 Well
- 48 How well are you able to do your language assignments in class?
 Not well at all
 Not well
 Not so well
 Well
- 49 How well are you able to remember what you have learned at school?
 Not well at all
 Not well
 Not so well
 Well
-
- 50 How well are you able to go to parties?
 Not well at all
 Not well
 Not so well
 Well
- 51 How well are you able to play with other children at school during recess?
 Not well at all
 Not well
 Not so well
 Well
- 52 How well are you able to play with other children after school?
 Not well at all
 Not well
 Not so well
 Well
- 53 How well does it go when you have dinner at a friends' house?
 Not well at all
 Not well
 Not so well
 Well
- 54 How well does it go when you visit someone, such as a family member or other children?
 Not well at all
 Not well
 Not so well
 Well

PART 3

The next questions are about whether it bothers you if things do not go well, for example, when you don't feel well or when you are sick.

Example

Does it bother you when you have trouble playing a certain sport?

If it does not bother you that playing a sport does not go well, then place an "x" on the second face just as it is done here below:



Not at all

Just a little

Quite a bit

A great deal

If you do not have trouble playing a certain sport, then try to imagine how it would feel if you had trouble with playing a sport.

Please answer what you think and try to answer each question.

55 Does it bother you when you have trouble biking?



Not at all



Just a little



Quite a bit



A great deal

56 Does it bother you when you have trouble playing outside?



Not at all



Just a little



Quite a bit



A great deal

57 Does it bother you when you have trouble running?



Not at all



Just a little



Quite a bit



A great deal

58 Does it bother you when you have trouble joining in gym?



Not at all



Just a little



Quite a bit



A great deal

59 Does it bother you when you have trouble swimming?



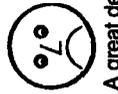
Not at all



Just a little



Quite a bit



A great deal

60 Does it bother you when you have trouble doing math assignments in class?



Not at all



Just a little



Quite a bit



A great deal

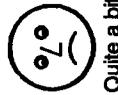
61 Does it bother you when you have trouble concentrating on your schoolwork?



Not at all



Just a little



Quite a bit



A great deal

62 Does it bother you when you have trouble doing language assignments during class?



Not at all



Just a little



Quite a bit



A great deal

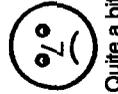
63 Does it bother you when you can't remember what you learned at school?



Not at all



Just a little



Quite a bit



A great deal

64 Does it bother you when you are not able to go to a party?



Not at all



Just a little



Quite a bit



A great deal

65 Does it bother you when playing with other children during recess does not go well?



Not at all



Just a little



Quite a bit



A great deal

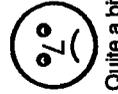
66 Does it bother you when playing with other children after school does not go well?



Not at all



Just a little



Quite a bit



A great deal

67 Does it bother you when you are not able to have dinner at a friend's house?



Not at all



Just a little



Quite a bit



A great deal

68 Does it bother you when you are not able to pay a visit to someone, such as a family member or other children?



Not at all



Just a little



Quite a bit



A great deal

69 Does it bother you when you have a headache?



Not at all



Just a little



Quite a bit



A great deal

72 Does it bother you when you get up feeling tired?



Not at all



Just a little



Quite a bit



A great deal

73 Does it bother you when you have to cough?



Not at all



Just a little



Quite a bit



A great deal

74 Does it bother you when you are short of breath?



Not at all



Just a little



Quite a bit



A great deal

75 Does it bother you when you are wheezy?



Not at all



Just a little



Quite a bit



A great deal

76 Does it bother you when you wake up at night due to your asthma



Not at all



Just a little



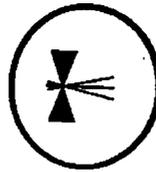
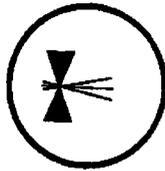
Quite a bit



A great deal

THANK YOU FOR COMPLETING THIS QUESTIONNAIRE!

If you think we left something out or forgot to mention something, then you can write it down in the space below. This can be about your illness or things that you do or have experienced. You are also allowed to make a drawing.



APPENDIX 3 – CHILD ATTITUDE TOWARD ILLNESS SCALE (CATIS)

How do you feel about having asthma?

Below are 12 questions concerning you and your feelings. Please read every question carefully. Answer each question, even though it may be difficult choosing only one answer. There are no right or wrong answers. Only you can tell us how you really feel.

Place an "x" in the circle that indicates your answer.

1. How often do you feel terrible about having asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

2. How often do you feel that it is unfair that you have asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

3. How often are you sad about having asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

4. How often do you think that it is your fault that you have asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

5. How often is it difficult to do the things you like to do because of your asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

6. How often do you think that you will always be ill?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

7. How often do you feel that your asthma keeps you from doing new things?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

8. How often do you feel different from others because of your asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

9. How often do you feel fed up with having asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

10. How often do you feel sad about having an illness?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

11. How often do you feel happy, even though you have asthma?

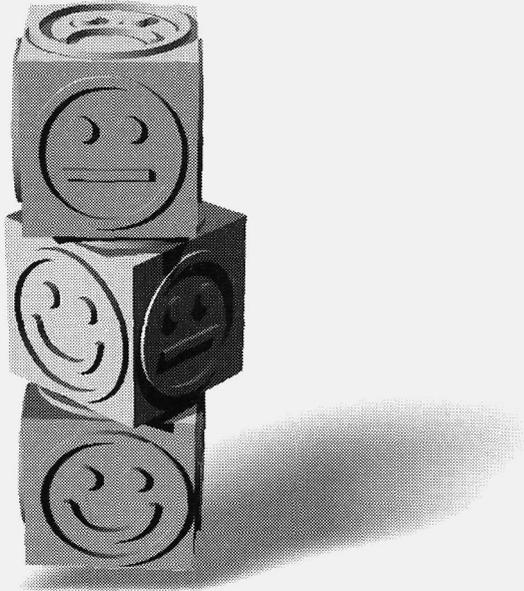
0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

12. How often do you feel as good as other children your age, even though you have asthma?

0	0	0	0	0
Never	Not often	Sometimes	Often	Very often

CURRICULUM VITAE

Jeanet Bruil was born in Gorssel (the Netherlands) on 22nd January 1963. In 1981 she passed the secondary school exam at the Baudartius College in Zutphen. In 1987 to 1988 she was a student assistant in the Leiden Institute for Social Policy Research. In May 1988 she took her master's degree in Special Education and Child Care at Leiden University. From 1989 to 1990 she was a part-time researcher at the Center for Science and Technology Studies at Leiden University and from 1989 to 1992 she was also attached as a part-time researcher at the Centre for Youth Welfare at Leiden University. From 1992 to 1997 she fulfilled her Ph.D. in the Clinical and Health Psychology Department at Leiden University. From 1998 tot 1999 she held an appointment as lecturer and postdoctoral researcher in the Department of Clinical and Health Psychology at Leiden University. Since October 1999 she has been working as a researcher at TNO Prevention and Health, Child Health Division.



Jeanet
Bruil

Development of a Quality of Life Instrument for Children with a Chronic Illness



Health-related quality of life is often used as a construct to define the broader spectrum of outcomes of illness and treatment and is becoming increasingly important in pediatrics. In this doctoral dissertation, health-related quality of life refers to the physical, social and psychological functioning of children and the gap between their achievements and expectations related to their functioning.



Health-related quality of life instruments can assist in structuring and transmitting clinical information, screening populations at risk, describing the quality of life of patient groups, and assisting in the evaluation of interventions. Until now, however, no health-related quality of life instruments were available for children that were generic as well as disease-specific, multi-dimensional, including child as well as parent report, including the level of functioning as well as related feelings, and assessing negative as well as positive aspects of health-related quality of life.



This study describes the development of the *How Are You?* (HAY), a health-related quality of life instrument for children aged 7 to 13 years. The HAY is one of the first instruments that meet all these criteria. In addition, it is the first study known to use confirmatory factor analysis using LISREL to confirm the hypothesized structure of a questionnaire which is a health-related quality of life instrument for children based on large groups of children with a chronic illness and healthy children.



Results reveal that the HAY is a valid and reliable instrument when used for research purposes. For clinical purposes, it can be used to guide the communication between patient and physician. Furthermore, it is easy to answer and parents and children approve of its use. It is recommended that children as well as their parents answer the questionnaire as both reports are bound to their own situation and evaluations.

*Jeanet Bruil is presently working as a researcher at
TNO Prevention and Health, The Netherlands*