Using health-related quality of life instruments for children with long-term conditions

On the basis of a national quality registry system

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School of Health and Welfare
Dissertation Series No. 77, 2017
Doctoral Thesis in Health and Care Sciences

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Dissertation Series No. 77

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Publisher
School of Health and Welfare
P.O. Box 1026
SE-551 11 Jönköping
Tel. +46 36 10 10 00
www.ju.se

Printed by Ineko AB 2016

ISSN 1654-3602
ISBN 978-91-85835-76-8
“The real voyage of discovery consists not in seeking new landscapes, but in having new eyes”

Marcel Proust
Abstract

Introduction: There has been a continuous development of new technologies in healthcare that are derived from national quality registries. However, this innovation needs to be translated into the workflow of healthcare delivery, to enable children with long-term conditions to get the best support possible to manage their health during everyday life. Since children living with long-term conditions experience different interference levels in their lives, healthcare professionals need to assess the impact of care on children’s day-to-day lives, as a complement to biomedical assessments. Knowledge about how health-related quality of life (HRQOL) instruments could be useful as valuable resources in clinical practice is needed. Therefore, it is important to explore the views of children and healthcare professional regarding such instruments and observe their role in the healthcare system.

Aim: The overall aim of this thesis was to explore and describe the use of instruments about health-related quality of life in outpatient care for children with long-term conditions on the basis of a national quality registry system.

Methods: The research was conducted by using comparative, cross-sectional and explorative designs and data collection was performed by using different methods. The questionnaire DISABKIDS Chronic Generic Measure -37 was used as well as semi-structured interviews and video-recordings from consultations. Altogether, 156 children (8–18 years) and nine healthcare professionals participated in the studies. Children with Type 1 Diabetes (T1D) (n 131) answered the questionnaire DISABKIDS and children with rheumatic diseases, kidney diseases and T1D (n 25) were interviewed after their consultation at the outpatient clinic after the web-DISABKIDS had been used. In total, nine healthcare professionals used the HRQOL instrument as an assessment tool during the encounters which was video-recorded (n 21). Quantitative deductive content analysis was used to describe content in different HRQOL instruments. Statistical
inference was used to analyse results from DISABKIDS and qualitative content analysis was used to analyse the interviews and video-recordings.

**Results:** The findings showed that based on a biopsychosocial perspective, both generic and disease-specific instruments should be used to gain a comprehensive evaluation of the child’s HRQOL. The DISABKIDS instrument is applicable when describing different aspects of health concerning children with T1D. When DISABKIDS was used in the encounters, children expressed positive experiences about sharing their results with the healthcare professional. It was discovered that different approaches led to different outcomes for the child when the healthcare professionals were using DISABKIDS during the encounter. When an instructing approach is used, the child’s ability to learn more about their health and how to improve their health is limited. When an inviting or engaging approach is used by the professional, the child may become more involved during the conversations.

**Conclusions:** It could be argued that instruments of HRQOL could be used as a complement to biomedical variables, to promote a biopsychosocial perspective on the child’s health. According to the children in this thesis, feedback on their results after answering to web-DISABKIDS is important, which implies that healthcare professionals need to prioritize time for discussions about results from HRQOL instruments in the encounters. If healthcare professionals involve the child in the discussion of the results of the HRQOL, misinterpreted answers could be corrected during the conversation. Concurrently, this claims that healthcare professionals invite and engage the child.

**Keywords:** outpatient setting; patient perspectives; patient reported outcomes; biopsychosocial perspective; self-reports; child involvement; International Classification of Functioning Disability and Health version for Children and Youth
Collection of papers

This thesis is based on the following papers which are referred to by their Roman numerals in the text.

Paper I


Paper II


Paper III


Paper IV


The articles have been reprinted with the kind of permission of the respective journals.
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Introduction

Historically, the healthcare system has been built from a provider perspective, but during the last two decades, the demands on improvement of the quality of care have been addressed by elucidating the individual perspective (Bergman, Hellström, Lifvergren, & Gustavsson, 2015). Reports coming directly from the person about their health condition in combination with clinical data, could gain this perspective (Carlier et al., 2012). One example is patient reported outcomes (PRO) which can be used at a personal level to improve interactions between the child and the professional (Snyder, Jensen, Segal, & Wu, 2013), or to improve the discussion and detection of health-related quality of life (HRQOL) problems (Greenhalgh, 2009). However, there is a question of exploring how PROs might address children’s and professionals’ collaborative use of these measures and observations about their role in the healthcare system.

To date, there has been a continuous development of new technologies and knowledge in healthcare derived from national quality registries (Ovretveit et al., 2013). However, this collection of knowledge and innovation needs to be translated into the workflow of healthcare delivery, to enable children and their families to get the best support they can get to manage their everyday life. It is stated in the United Nations Convention on the Rights of the Child (UNCRC) that every child should have equal rights and the right to live as normal life as possible regardless of their prerequisites (UNCRC, 1989). Consequently, children should be consulted on their own goals of treatment to maximize their participation in the encounter, which also should be an overarching goal in healthcare (Morris, 2009). Children who have a long-term condition are facing different challenges in their everyday life due to their long-term condition. Children with kidney diseases have described concerns about their quality of life concerning school functioning and depressive symptoms (Gerson et al., 2010; Kogon et al., 2016), and children with Juvenile Idiopathic Arthritis (JIA) concerning their physical functioning (Lundberg, Lindh, Eriksson, Petersen, & Eurenius, 2012). Children with other long-term conditions such as asthma, cystic fibrosis and diabetes have also reported their
HRQOL as worse than healthy children, especially regarding the participation in physical activities and in family activities (Sawyer et al., 2004).

It has been found that in situations when healthcare professionals have access to information about the child’s HRQOL, they are more likely to discuss these issues with the child (Marshall, Haywood, & Fitzpatrick, 2006). Since children living with long-term conditions experience different levels of interference in their lives, healthcare professionals need to assess the impact of care on children’s day-to-day lives, as a complement to biomedical assessments (Corathers et al., 2015). The reports from children can provide insights that could help professionals with difficult decisions (Fayed et al., 2012), although research has found that children’s participation in conversations in encounters tend to be rather scarce (Coyne, 2008; Wassmer et al., 2004). Hence, this thesis aims to consider the potential use of instruments about health-related quality of life in outpatient care for children with long-term conditions, on the basis of a national quality registry system.
Background

Children with long-term conditions

The concepts chronic illness, chronic health condition, medical disease or long-term condition is often used differently in the literature. The definitions of long-term conditions exhibit heterogeneity in characteristics such as duration, need for medical treatments, risk factors and effect on well-being (Goodman, Posner, Huang, Parekh, & Koh, 2013). In general, a long-term condition is characterized by three features; the prolonged duration of more than three months; the condition cannot resolve spontaneously; and the condition is rarely cured completely (Compas, Jaser, Dunn, & Rodriguez, 2012; van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007).

The prevalence of children with long-term conditions vary, dependent on methods and definitions, however, the estimation is probably underestimated since the increase in obesity is driving an increase in other long-term conditions concerning children (Van Cleave, Gortmaker, & Perrin, 2010). In the United States, epidemiological studies suggest that about 25 % of children in the aged 0–17 years suffer from a long-term condition (Van Cleave et al., 2010; van der Lee et al., 2007). Thus, this number is not self-reported from children themselves. In Sweden, there are about 20 % of children in the aged 13–15 years that have described themselves as having a long-term condition which affects their daily life (Folkhälsomyndigheten, 2014). Furthermore, there are disparities in self-reported health between children who have a condition compared to the healthy ones (Statens folkhälsoinstitut, 2012). Moreover, it has been described that children with long-term conditions experience more stress-related symptoms and more mental health problems in their daily life, compared to their healthy counterparts (Folkhälsomyndigheten, 2014; Hysing, Elgen, Gillberg, & Lundervold, 2009; Statens folkhälsoinstitut, 2012). Since long-term conditions are associated with high rates of emotional, developmental and behavioural problems, more focus should be on sorting out the complex interactions between severity and control of the disease, and socio-environmental factors (Blackman, Gurka, Gurka, &
Unarguably, diagnoses of long-term conditions are usually stable, but functioning does tend to change over time, which requires attention on the functional needs of these children (Lollar, Hartzell, & Evans, 2012). In this thesis, three different kinds of long-term conditions are in focus, and a brief description is provided of each disease.

**Long-term conditions related to this thesis**

Type I diabetes (T1D) is an autoimmune condition which is caused by the destruction of the insulin producing cells in the islets of Langerhans in pancreas. This leads to insulin deficiency which causes high blood glucose values and symptoms such as fatigue and increased weight loss (American Diabetes Association, 2005). The cause of T1D is still mostly unknown and in children it affects their lives as they need daily medical treatments with injections or infusions of insulin, and regular blood glucose monitoring for adapting insulin doses etc. (Silverstein et al., 2005).

Juvenile idiopathic arthritis (JIA) is a broad term that describes a heterogeneous group of arthritis with unknown causes. The term encompasses several disease categories with different clinical signs and symptoms such as swollen joints, pain and fatigue, which affects the daily life of children who suffer from the condition. The cause is still poorly understood but seems related to genetic and environmental factors. None of the available drugs for treating JIA are curative, but the prognosis has been greatly improved as a result of the development of new biological therapies (Ravelli & Martini, 2007).

Chronic kidney disease (CKD) refers to a condition related to the irreversible damage of the kidneys. The progression of CKD varies and is dependent on the underlying disease or initial injury as well as additional risk factors. Some examples of underlying causes are congenital defects (malformations of kidney or urinary tract), hereditary nephropathies and glomerulonephritis, and each cause varies as the child gets older. CKD is associated with hypertension, proteinuria and anaemia (Harambat, Van Stralen, Kim, & Tizard, 2012) and can affect the children concerning their emotional functioning and school work (Neul, Minard, Currier, & Goldstein, 2013).
An effective management of children’s long-term conditions requires that children and their family receive appropriate clinical care, with support to manage their everyday life. Positive outcomes are more likely if the person can seek and receive help and if the healthcare professionals communicate effectively by developing a shared understanding about the problem (Batalden et al., 2015).

Children in the healthcare system

Unarguably, children who need support from the healthcare system shall have their right to express their opinions, which is stated in article 12 in the UNCRC. In addition, all children who have some kind of physical or psychological disability shall have the right to live an adequate and decent life, which enables active participation in society (article 23) (United Nations, 1989). Moreover, the Patient Health and Medical Services Act (SFS 2014:821) points out that healthcare shall be designed in joint consultation with the patient and that patients shall participate in their own care and/or treatment according to their individual circumstances (chapter five §1-2) which also counts for children (SFS 2014:821). With this point in mind, studying how children share their experience of living with a long-term condition to healthcare professionals is important. By asking children about their own perspective, it could be argued that healthcare professionals could learn more about how to support the individual child (Johansson, 2003). Children have described that they appreciate the qualities of honesty, good listening and confidentiality in the consultation, which is more important than meeting a professional with the same gender or ethnicity (Farrant & Watson, 2004). Still, the evidence describes many obstacles to the realisation of children’s participation in the healthcare setting. Despite the large volume of research about the support of children’s rights to be listened to, the actual implementation in practice is questionable (Coyne, 2008). It has been demonstrated that healthcare professionals support the inclusion of children and consider the child’s preferences for participation in decisions and treatments (Vaknin & Zisk-Rony, 2011). However, children have described that routine consultations are often boring and that the professionals are taking the lead and setting the agenda. The consultations give little space for the child to discuss their
concerns, since parents and professionals do most of the talking, although the child is capable of playing a more active role. It is argued that the current structure of healthcare consultations and communication styles employed may hinder children’s involvement during consultations (van Staa, 2011). Certainly, there is a gap between ideology and practice about involving children in their healthcare, and the healthcare system is still having difficulties in putting the child first (Bate & Robert, 2006). Therefore, an understanding of how the healthcare system is organised that goes beyond cost-efficiencies to coordinate care around services (Goodwin, 2016) should be explained.

The healthcare system

According to the Chronic Care Model (CCM), which is as an evidence-based model, the healthcare system is described as part of a larger community (Wagner et al., 2001; Wagner, Davis, Schaefer, Von Korff, & Austin, 1999). The management of long-term conditions needs to be linked with available resources in the broader community. Leadership, incentives and self-management support can promote the development of informed, active patients and proactive prepared professional teams. It is the interaction between the patient and the team that will assure delivery of services and promoting outcomes. Being prepared means having the necessary expertise, time, resources and information to assure clinical management. The patient must have the confidence and information to make the best use of their involvement together with the team (Wagner et al., 1999). A healthcare system where professionals and patients interact as participants within the system at different levels should be a general goal. At the basic level, this requires civil discourse dependent on the respectful interaction and effective communication between patients and professionals (Batalden et al., 2015), which means that a clinical information system is a critical feature for effective care. By providing healthcare professionals with access to such systems, it is possible to identify special needs, deliver planned care and receive feedback about results (Wagner et al., 1999). The healthcare of today should be driven by more requirements, which have been identified for better quality in healthcare.
Quality in the healthcare system

When describing the quality of healthcare, the National Board of Health and Welfare have outlined areas such as safety, timeliness, efficiency, equity and patient-centeredness. This implies that the care should be built on the respect for each person’s needs which shall be considered when clinical decisions are made (Socialstyrelsen, 2006). Consequently, quality in healthcare needs to be valued by the one who is going to participate in the care. This means that the patient (child and/or parents) should have the opportunity to describe their needs and expectations. Other ingredients also described by Bergman and Klefsjö (2010), are the processes to create value, which means that networks are needed that include healthcare professionals, patients and others in society. In those networks, process views can facilitate the linking of data from the past to present, which in turn can be helpful when identifying areas for improvement. Innovative ways to better perform are also needed, which requires a visionary leadership that supports the healthcare professionals in their improvement work. It is also necessary to gather information, to make the right decisions based on facts (Figure 1). To do this, knowledge about variation is important and data should be gathered in a structured way and consists of different types of information (Bergman et al., 2015; Bergman & Klefsjö, 2010), such as professional-based measures as well as patient reported outcome measures. Consequently, data collection and monitoring in feedback systems are key aspects to improve quality (Dixon-Woods, McNicol, & Martin, 2012) and therefore a national quality registry could be exemplified as one system for monitoring the quality of healthcare.
Figure 1. Principles for quality in healthcare, figure adapted and abbreviated from Bergman & Klefsjö (2010) and Bergman et al (2015)
National Quality Registry systems

A national quality registry can facilitate learning and continuous improvements and is seen as a necessary ingredient of a modern healthcare system in Sweden (Jacobsson Ekman, Lindahl, & Nordin, 2016). Most of the registries are created by healthcare professionals and are utilized by the departments in Sweden. Furthermore, these registries contain data about patient problems/diagnoses, medical interventions and outcomes, and are standardized which provide possibilities to follow up achievements in healthcare as well as a way to improve healthcare (SKL., 2009). The registries are fostered by patient data regulation and an indirect control by a combination of government funding and professional self-governance (Levay, 2015). One vision for the registry is to be an overall support that could actively be used at different levels of learning, improvement and governance of healthcare activities (Jacobsson Ekman et al., 2016). Since the registries are national in their coverage, comparisons can be made between county councils and departments and monitored over a period of time. The comparisons of data from a registry can be used to guide or evaluate improvement efforts, however, this will not imply that a registry will automatically yield better care and active efforts based on the knowledge of what needs to be changed and how to achieve this, have to be included (Jacobsson Ekman et al., 2016).

The quality registry system in this thesis

There are different levels in the quality registry system. The regulations for quality registries are built on group-comparisons and therefore, the individual level (used in this thesis) is based on a pre-system to the quality registry as described in Figure 2. The national quality registry for adults with rheumatoid arthritis (SRQ) has served as a role model when the feed-forward system (FFS) was developed. The FFS is integrated into the process of care delivery. Children enter data about their health-related concerns prior to the planned consultation at the clinic and it is then downloaded into the system. Previously entered data is compiled into a summary page (dashboard) with graphical representations of the child’s health status followed over time. This page also includes treatments obtained from the electronic medical record (EMR), entered by the
healthcare professional and the summary page can be used as a decision support tool during the encounter (Hvitfeldt et al., 2009). The system consists of three parts, with separated data storage (Figure 2). The first part is the platform for personal measures (PM), where children reply to different questionnaires, handled by the child with individual credentials. The second part is a display platform for the decision support (DS) where all inserted data is visualised. This part is handled by the healthcare professional. The third part is the national quality registry which is for data storage for group comparisons.

The regulations demand that healthcare professionals fully inform patients about the aim of the national quality registry and the rights to opt-out. The information requirements are the same as the requirements for opt-in, however, the only difference is that the patient needs to explicitly refuse to participate in the national quality registry (Gliklich, Dreyer, & Leavy, 2014). The Patient Data Act (SFS 2008:355) describes how the information exchange shall be organised to provide for patient safety, high quality and cost-effectiveness. The information exchanges shall also be built on respect for each individual integrity and this holds true for both the medical health record as well as for the quality registry (SFS 2008:355).
Figure 2. Flowchart describing the feed-forward system preceding the registry (PM: personal measures; DS: Decision support; EMR: Electronic Medical Record)

From a wider perspective, the registries are generally limited in regards to following the patient perspective throughout the care cycle since they are designed mainly to be used by professionals and is focusing on specific diseases or interventions. Therefore, the incorporation of patient reported outcomes and collaboration between registries are needed (Levay, 2015). For learning and improving quality, medical assessments need to be supplemented with the child’s own assessment of health and wellbeing (Jacobsson Ekman et al., 2016). Therefore, a more detailed description of these concepts is outlined below.
Health and quality of life

A well-known definition of health is provided by the World Health Organization (WHO) as follows: "Health is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity" p 459 (World Health Organization, 1958). The main point in this definition is the holistic perspective with the inclusion of social, mental and physical well-being. However, the absoluteness of the word “complete” would leave most people being regarded as unhealthy (Huber et al., 2011). Therefore, another definition proposing the holistic approach can be described, which is called the welfare theory of health. Health is considered to be a primary concept in the web of medical concepts. A person has good health if he or she is in that kind of a physical and mental condition that he or she can realise his or her vital goals in any given circumstance (Nordenfelt, 2004). In this perspective, a child with a long-term condition can be in good health. Health could also be described as the ability to carry out activities which are essential in the person’s life. The person is creating his or her own reality in interaction with others which includes the creation of health with focus on the whole person, the family and community (Parse, 1990).

Since the WHO statement of health in the late 1950s, it has been raising expectations about health. This has led to a shift to emphasise a person’s capacity and ability to perform daily activities, emotional well-being and quality of life (QOL). For the individual, health changes over the course of their life and involves their own assessment of well-being as well as the views of others and society as a whole (Callahan, 2000; Lindau, Laumann, Levinson, & Waite, 2003). The concept of QOL has become widely used as an indicator when evaluating treatments. QOL could be seen as personal and unique, without any general understanding and it is vague and something people can talk about, but which few can define (Bowling, 2005). In research, there has been a growing interest of using QOL as an outcome measure. In general, QOL refers to the goodness of those aspects that could affect health, but are influenced by many other factors than health, such as job satisfaction, income, environment and social opportunities. In other words, QOL covers all aspects of life including health status, environment, financial aspects and human rights. QOL
means different things to different people, and can take different meanings according to the area of application (Bowling, 2005). The concept of QOL usually includes a person’s subjective evaluations of both positive and negative aspects of life, however, aspects of culture, values, and spirituality are also key aspects that adds to the complexity of the understanding of QOL (WHOQOL Group, 1995). There has been an increasing attention in research about QOL, however, there is still a lack of a clear conceptualization of QOL, and there is a challenge for the research society to take a stand for a clearer definition (Moons, Budts, & De Geest, 2006).

**Health-related quality of life**

To encompass the overall aspects of QOL that can affect health, the term health-related quality of life (HRQOL) has been introduced. HRQOL excludes aspects of QOL that are not related to health, for example cultural, political or societal attributes (Ferrans, Zerwic, Wilbur, & Larson, 2005). On the individual level, HRQOL can include both physical, mental and social health perceptions and the correlations to a condition (disease or illness), functional status, social support, health risks and socioeconomic status (Cramer & Spilker, 1998). The concept of HRQOL is embracing functioning, well-being and feelings about health, for example functional status, which refers to what the child actually can do, whereas QOL is referenced to how the child feels (E. Davies et al., 2006). There is no evidence that the child’s perception of his or her life is corresponding to the ability to perform activities, therefore, it is possible that children with poor functioning can have high HRQOL. The common sense is that HRQOL is a multidimensional construct, which is seen as a subjective description of well-being and functioning (E. Davies et al., 2006). According to the WHO’s definition, HRQOL emphasises the personal perception of the quality of one’s life based on personal expectations. The uniqueness of a person’s own experience of how the condition emerges, suggest that two people with the same disease can judge their HRQOL very differently (Ferrans et al., 2005).
Biopsychosocial models of health and illness

It has been argued that the individual perspective of disease and treatment, social context and the complementary system devised by the healthcare system is important, therefore a biopsychosocial model could be used. Engel’s (1977) biopsychosocial model of health has its roots in the general system theory which have been developed by von Bertalanffy (1968). The argument was the need for a fundamental reorientation in the scientific perspective in order to open a way for a holistic approach. System theory posits that several levels of organisation are linked in a hierarchical relationship and a change in one level effects a change in others (Von Bertalanffy, 1968). Indeed, the study of a disease and the resulting medical care shall be seen as interrelated processes (Engel, 1977). The biopsychosocial model introduced the clinical importance of interacting psychosocial factors in contrast to the biomedical model. The biopsychosocial model is a descriptive framework that permits the exploration of the person’s experience of illness to yield a comprehensive picture of the health condition. In order to understand disease, it is important to consider the persons thoughts, experiences, abilities as well as the social context in the interaction with biological processes (Engel, 1977): “The boundaries between health and disease, between well and sick, are far from clear and never will be clear, for they are diffused by cultural, social, and psychological considerations.” p 132 (Engel, 1977). This perspective on health provides a reference on how to apply a holistic approach and to expand to the contextual needs of each person.

A further description of the biopsychosocial perspective is the International Classification of Functioning and Disability (ICF), which reinforces Engel’s model. ICF integrates the social perspectives of activities and participation into a clinical understanding (Costa-Black, Feuerstein, & Loisel, 2013). In the ICF model, health is described according to three different levels; the body, the individual and the society. ICF-CY (for children and youth) is a conceptual framework that provides terminology for recording functions and structures of the body, activity and participation and relevant environmental factors manifested in childhood. The structure of ICF-CY is a theoretical model and a
classification system which provides a standard language and framework for descriptions of health, health-related states and well-being, for applications to various aspects of health (World Health Organization, 2007). The ICF model attempts to provide a coherent view of health from biological, individual and social perspectives. The theoretical model of ICF consist of six components. The *health condition* and its relation to *body functions and structures*, which is the functioning of bodily organs and systems. The *activity* refers to the execution of a task or actions made, and *participation* refers to involvement in a life situation, for example interaction with peers. The two contextual components are *environmental factors* and *personal factors* which belong to the context of the person’s life. Environmental factors have an impact on the other components and are organised from the persons’ immediate environment to a general environment. The personal factors provide information about age, gender or social status, which is not classified by codes in ICF-CY (World Health Organization, 2007).

In conclusion, the person’s own values and preferences are likely to affect the experiences of health and illness (Ferrans et al., 2005). Having a long-term condition as a young person can have several impacts on well-being as described earlier. Healthcare professionals should be aware of the wider psychosocial issues that could be important to children with long-term conditions (Farrant & Watson, 2004). One way to do this is by using instruments for measuring HRQOL.

**Measuring HRQOL in children**

When measuring HRQOL the person is the expert (Morrow, Quine, Heaton, & Craig, 2010). A subjective assessment of the persons’ experiences of health, symptoms, functioning, well-being, HRQOL or perceptions about treatment and satisfaction with the care received is often called patient reported outcomes (PRO). It can be a summary of a personal evaluation of a disease and treatment which provides perceptions related to the condition and its impact on daily life (Rothman, Beltran, Cappelleri, Lipscomb, & Teschendorf, 2007). PRO provides information that is unavailable from other sources and reflects how a person interprets the experience of having a condition, which is not observable by others.
(Rothman et al., 2007). PRO should be answered by the person alone, without interpretation from others, for example healthcare professionals (Valderas & Alonso, 2008) or parents. Indeed, there are differences between how parents, children and professionals rate HRQOL (Morrow, Hayen, Quine, Scheinberg, & Craig, 2012), therefore it is important to ask children themselves as far as possible, since children’s perspectives tend to relate to "here and now", whereas parental perspectives often reflect nuanced and abstract elements, such as elements of the future (Nicholas et al., 2010).

It has been emphasised that children’s knowledge about health and illness becomes more accurate and sophisticated with age. Children’s own experiences about health and illness is often described in terms of presence of symptoms or feeling poorly. Moreover, children describe health based on behavioural factors, and not necessarily as opposite to illness (Myant & Williams, 2005). It is also known that children have considerable knowledge about health and illness and their attitude is that health could be an act as a positive resource in life with a strong holistic view on health, which also have been described by adults (Piko & Bak, 2006). According to children, emotional and mood elements are associated with well-being, i.e. sadness, joy or a sense of freedom (Davó-Blanes & La Parra, 2013). Therefore, children are capable of providing relevant information about their health and illness. It has been found that a range of issues about health are important to discuss from the child’s perspective (A. Davies & Randall, 2015; Farrant & Watson, 2004), which highlights the importance of making children’s own descriptions of health and well-being noticeable.

**The use of HRQOL instruments**

Why is it interesting to use HRQOL instruments in clinical practice? Firstly, improving HRQOL for children living with long-term conditions has value in itself, since it has connections to the psychological aspects of the disease. HRQOL instruments can also be used to evaluate treatment effects and as an outcome measure. In addition, communicating the individual results can provide professionals with insights into the child’s experiences of symptoms and functioning (Lavallee et al., 2016).
There are instruments that are intended to assess situations across different health conditions and across different cultures. These instruments are generally called generic instruments. The consequence is that these instruments are not sensitive to small changes, therefore disease-specific instruments can be added to assess disease-specific functions (Harding, 2001). There has been a dramatic increase in the use of HRQOL instruments to evaluate patient outcomes as well as treatments which reflect an emerging shift in healthcare. A systematic review has shown that PRO could be effective when it is used as a management tool for adult patients in outpatient care and concerning a specific population (Boyce & Browne, 2013). A few studies have been conducted regarding the use of PRO in paediatric care. Positive results were shown on the effects on psychosocial well-being in adolescents with diabetes when HRQOL results were discussed (de Wit et al., 2008). The assessment of electronic HRQOL profiles in children with JIA have demonstrated increased discussion of psychosocial functioning (Haverman et al., 2013) as well as in paediatric oncology (Engelen, Detmar, et al., 2012). Results showed that the use of a HRQOL instrument increased the number of psychosocial topics, but found that it does not influence the type of psychosocial problems during the consultation. This study found that the use of PRO has a positive impact on the communication between the child and the physician. Further, the use of PRO may help the healthcare professional to identify psychosocial problems, but the intention was not to solve the problems (Engelen, van Zwieten, et al., 2012). It has also been stated that the use of HRQOL instruments may improve the efficacy of the consultation as the healthcare professional is better equipped to focus quickly on current HRQOL problems and sub-domains (Engelen, Detmar, et al., 2012; Haverman et al., 2013).

It could be concluded that repeated HRQOL assessments may provide useful information concerning the child’s adaptation to their condition. However, a lack of time and expertise in interpreting and scoring of the HRQOL data in clinical practice is one obstacle (Abbott, 2009). An important strategy for routine assessment of HRQOL is the need to help healthcare professionals understand the importance of evaluating HRQOL as well as the benefits in terms of children’s well-being and resource allocation. Involving the child can help assure that the proposals are
acceptable and feasible to support them when the use of the HRQOL instrument is introduced (Wray, Brown, Marino, & Franklin, 2011). If the results of the HRQOL instruments shall be accessible, there is a need for web-based designs of these instruments and to educate the healthcare professionals who are using the instruments as well as optimising the effect on using PROs in a clinical context is also crucial (Haverman, Engelen, van Rossum, Heymans, & Grootenhuis, 2011). Hence, the focus on children’s attitudes towards answering an HRQOL instrument prior to the consultation should be explored. Qualitative studies on how professionals and children make sense of HRQOL instruments in clinical practice also needs to be considered (Greenhalgh, 2009).
Rationale

Healthcare has focused primarily on reducing mortality and morbidity in terms of survival and hospitalizations for children with long-term conditions. However, outcomes focusing on the impact of the disease in everyday life and social functioning may be as important as the symptoms of the disease. Children have described that they desire to share information about their HRQOL, although healthcare professionals do not always ask about these concerns in a systematic way. Children can also have difficulties expressing feelings and other issues concerning their health during consultations. PRO are a core aspect of healthcare, but there is a challenge to use such instruments in clinical practice to improve the care for children with long-term conditions. Reports coming from the child can provide important information from the child’s perspective and can be used to evaluate disease progress and treatment effects on the child’s everyday life. Moreover, it could help healthcare professionals with targeted interventions that may improve care. There has been an increase in the use of HRQOL instruments to evaluate patient outcomes as well as treatments, towards valuing the perspective from each person. However, few studies have evaluated the use of HRQOL instruments in a clinical practice setting. Clinical interventions are needed to provide knowledge about how HRQOL instruments could be useful as valuable resources in clinical practice. Therefore, it is important to explore the views of children and healthcare professional regarding such instruments and observe their role in the healthcare system as a whole.
Aim

The overall aim of the thesis was to explore and describe the use of instruments about health-related quality of life in outpatient care for children with long-term conditions on the basis of the national quality registry system.

Specific aims of each paper

- To compare content dimensions of HRQOL instruments for children’s self-reports using the framework of ICF-CY (paper I).
- To describe differences in the HRQOL of children with T1D according to age, gender and metabolic control (paper II).
- To explore children’s experiences about a structured assessment of HRQOL (paper III).
- To explore how healthcare professionals use a structured assessment tool for HRQOL during paediatric encounters (paper IV).

Specific aims in thesis frame

- To describe the HRQOL among children with T1D according to a biopsychosocial perspective.
- To describe and compare children’s and healthcare professionals’ use of a HRQOL instrument in the patient encounter.
Materials and methods

Scientific perspective

The approach to research is based on assumptions related to the nature of reality, the relationship of the researcher to that being studied and the process of research (Creswell, 2012). The world is complex and there are no simple explanations; rather there are multiple factors coming together that interact in a complex manner. Research aims to capture some of this complexity, and therefore, an understanding of participant experiences and multiple perspectives are needed. Concepts and theories are constructed in research, which attempt to explain and make sense out of experiences described by those participating in the research (Corbin & Strauss, 2008). In that sense, pragmatism is allowing researchers to note utility aspects, which calls for reflexive research practice. The phenomena that are being studied can have different layers, and the question is about how to observe or measure these layers (Feilzer, 2010). A pragmatic approach places its emphasis on joint actions and shared meanings, which could be described as the inquiries undertaken to assess the workability of potential lines of actions, or the base for claimed assertions (Morgan, 2007). Research questions in this thesis have been phrased in discussions with healthcare professionals working with children with different kinds of long-term conditions. Several pathways can lead towards developing knowledge and understanding of the studied area because inductive and deductive reasoning can generate different perspectives, which is based on different methodological approaches. The deductive approach starts with models or theories to describe predictions or explanations that are observed. The inductive approach is derived from facts that have been acquired through observations (Chalmers, 2013). This thesis comprises both deductive and inductive methodologies that can generate knowledge from different approaches.
Methodological framework

Design

The studies in this thesis have comparative, descriptive, cross-sectional and explorative designs and different types of data collection methods have been used. The four papers which constitute this thesis are built on each other. The first paper was the starting point in which the HRQOL instrument was selected. This study had a comparative and descriptive design to describe the content dimensions of different HRQOL instruments. A comparative design is used when the purpose is to explain and analyse differences and similarities (Brink & Wood, 1998). In the second paper, a cross-sectional, descriptive and comparative design was selected to investigate HRQOL of children with one long-term condition: type 1 diabetes. The intent was to make inferences about the studied population at a single point in time (Kirkwood & Sterne, 2003). Descriptive investigations are a way to observe, count and classify the studied area by comparing groups (Polit & Beck, 2013). In paper III–IV an explorative design was selected to explore different aspects from children and healthcare professionals. Qualitative research could be used to describe and interpret experiences to provide new insights about a studied area. Explorative designs intend to explore research questions rather than offer conclusive solutions and are conducted in order to investigate a new area and to shed light on various ways things could be explored (Polit & Beck, 2013). Explorative research forms the basis of more conclusive research, and may result in alternative options to guide further research (Bell, 2014). An overview of each paper in this thesis is described in Table 1.
Table 1. Overview of each paper in the thesis

<table>
<thead>
<tr>
<th>Paper</th>
<th>I</th>
<th>II</th>
<th>III</th>
<th>IV</th>
</tr>
</thead>
<tbody>
<tr>
<td>Design</td>
<td>Descriptive, Comparative</td>
<td>Cross-sectional</td>
<td>Explorative</td>
<td>Explorative</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Descriptive, Comparative</td>
<td>Descriptive</td>
<td>Descriptive</td>
</tr>
<tr>
<td>Sample</td>
<td>Total sample of six instruments</td>
<td>131 children (8-18 years old) with T1D</td>
<td>25 children (10-17 years old) with T1D,</td>
<td>21 video recordings from nine different</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>rheumatic diseases, kidney diseases</td>
<td>professionals’ consultations</td>
</tr>
<tr>
<td>Data</td>
<td>Instruments (CHQ-CF, Kid-KINDL-</td>
<td>DCGM-37 and DM-10, data from SWEDIAB-</td>
<td>Semi-structured interviews</td>
<td>Audio videotaped recorded conversations</td>
</tr>
<tr>
<td>collection</td>
<td>R, EQ5D-Y, DCGM-37, KIDSCREEN</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>-52, PedsQL)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Data</td>
<td>Quantitative deductive content</td>
<td>Non-parametric tests, inferential</td>
<td>Qualitative inductive content analysis</td>
<td>Qualitative inductive content analysis</td>
</tr>
<tr>
<td>analysis</td>
<td>analysis; ICF-CY</td>
<td>statistics</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
**Settings**

In Sweden, children with long-term conditions receive their specialist care from outpatient paediatric clinics situated at paediatric departments at county, regional and university hospitals around the country. In total, there are 35 paediatric departments in Sweden. In paper II, ten outpatient paediatric clinics from different parts of Sweden were approached during an improvement collaboration initiated by the registry for children with T1D (SWEDIABKIDS). During this collaboration, the topic of distribution of the questionnaire DISABKIDS was discussed and presented.

The registries for children with rheumatic diseases and kidney diseases had started improvement projects during 2013 in order to begin with PRO measures in a clinical setting. In accordance with these registries, a clinical intervention was performed based on the information technology system with a feed-forward system (FFS). This included children’s input of data and response-giving from healthcare professionals that was built on the DISABKIDS questionnaire. The clinical intervention was performed at those outpatient departments that actively used their registry. Four separate paediatric outpatient clinics from two regional hospitals and one university hospital in the southern region of Sweden participated (paper III–IV). A short description of the three national quality registries related to this thesis is provided in Table 2.
Table 2. Short description of each national quality registry related to this thesis

<table>
<thead>
<tr>
<th>Registry</th>
<th>Started</th>
<th>Aim</th>
<th>Coverage</th>
</tr>
</thead>
<tbody>
<tr>
<td>SWEDIABKIDS</td>
<td>2000</td>
<td>To describe population based medical results and quality indicators in the care of children with diabetes in Sweden.</td>
<td>98 %</td>
</tr>
<tr>
<td>Swedish Paediatric Nephrology Registry</td>
<td>2011</td>
<td>To find etiological relationships and prognostic factors as well as evaluate treatment protocols to provide optimal health and quality of life for children with chronic kidney diseases.</td>
<td>60 %</td>
</tr>
<tr>
<td>Swedish Paediatric Rheumatology Registry</td>
<td>2009</td>
<td>To collaborate with healthcare professionals and patients to improve quality, safety and development of care to improve the overall quality of life in children with rheumatic diseases.</td>
<td>60 %</td>
</tr>
</tbody>
</table>

**Data collection**

Different methods for collecting data have been applied, which took place during the period from year 2011 to 2015. Data have been collected in three studies. The first study was carried out in 2011 when the instruments of HRQOL were collected (paper I). The second study was conducted in 2013 with the use of the questionnaire DISABKIDS and other measures from the registry for children with diabetes (SWEDIABKIDS) (paper II). The clinical intervention was performed in 2013, and a third study was conducted during 2014-2015 (paper III–IV).
Participants and criterions for inclusion

In the first paper, a total sample of six HRQOL instruments was chosen and the specific inclusion criterion was decided upon prior to the study. All the instruments under consideration for inclusion were self-reports, translations to the Swedish with proven psychometric properties. The mode of operation to be measured by the instrument was either health, quality of life or health-related quality of life. The instruments available in the registries for children with long-term conditions was another inclusion criterion. Only generic instruments and self-reports for children in the age interval from 5–18 years old were chosen. Proxy-reports were excluded, and if short versions had been developed, only the long version was included.

In the second paper, the inclusion criterions for participating were children—between 8–18 years old—diagnosed with T1D. Another criterion was that participants could speak and understand the Swedish language, since the questionnaire was in Swedish. Also, the children needed an e-mail address to receive the link to the questionnaire (Table 3). There were 204 children who agreed to participate and 131 questionnaires were returned, which produced a response rate at 64 %. Drop-outs were analysed by comparing age, gender body mass index standard deviation (BMI-SDS) and glycosylated haemoglobin (HbA1c) between responders and non-responders.

The inclusion criterion for participating in paper III was a condition with an estimated duration of at least three months according to the definition of a long-term condition (van der Lee et al., 2007). The additional selection criterions were various numbers of clinical visits/year and a variety in the duration of the condition. A convenient sample was used, as time points for data collection were prearranged. Those children already scheduled at the clinic on those days were given information about the study prior to the consultation. In total, 28 children were approached with different kinds of conditions (T1D, rheumatic diseases and kidney diseases). However, 25 children participated, since two children declined to participate and one did not fit the inclusion criterion (Table 3).

In the last paper (IV), the inclusion criterions for participating were that the healthcare professionals had attended an instructional session about web-DISABKIDS and had started to use the assessment tool in their
consultations prior to data collection (Table 3). Eleven healthcare professionals were asked to participate, but two declined due to a lack of time, therefore nine healthcare professionals participated. The participants were seven paediatricians and two registered nurses with a special education in paediatric nursing. All participants each had more than ten years of professional experience in the field. Twenty-three consultations were video-recorded, but two recordings failed due to technical problems, therefore, twenty-one consultations were used for data analysis in paper IV. The number of consultations ranged from one to six/participant.

Table 3. Demographic data of participants in paper II–IV

<table>
<thead>
<tr>
<th>Paper</th>
<th>II</th>
<th>III</th>
<th>IV</th>
</tr>
</thead>
<tbody>
<tr>
<td>Participants, children</td>
<td>131</td>
<td>25</td>
<td>21</td>
</tr>
<tr>
<td>Participants, professionals</td>
<td>-</td>
<td>-</td>
<td>9</td>
</tr>
<tr>
<td>Age</td>
<td>8-18 (mean 13)</td>
<td>10-17 (mean 13)</td>
<td>41-65 (mean 46)</td>
</tr>
<tr>
<td>Male</td>
<td>61</td>
<td>9</td>
<td>3</td>
</tr>
<tr>
<td>Female</td>
<td>70</td>
<td>16</td>
<td>6</td>
</tr>
<tr>
<td>T1D</td>
<td>131</td>
<td>8</td>
<td>6</td>
</tr>
<tr>
<td>Rheumatic diseases</td>
<td>-</td>
<td>11</td>
<td>9</td>
</tr>
<tr>
<td>Kidney diseases</td>
<td>-</td>
<td>6</td>
<td>6</td>
</tr>
</tbody>
</table>
Materials

First, a brief description of each instrument from the comparison in paper I is provided. Since one of these instruments is further used in this thesis, a more detailed explanation has been outlined.

Instruments (paper I)

**The Child Health Questionnaire – Child Form (CHQ-CF)**

This instrument intends to measure HRQOL which covers both physical and psychosocial domains and is widely used in many countries, both in the general population and in children with long-term conditions (Ruperto et al., 2001). The children’s self-report has subjects from 5–18 years of age. CHQ-CF is divided into ten subscales (physical functioning; role functioning; emotional, behavioural, physical; bodily pain; general behaviour; mental health; self-esteem; general health perceptions; change in health; family activities; and family cohesion). The 87 items are divided into four, five and six response options over a 10 multi-item scale and two single item scales. Most of the items have a recall period of four weeks, and the general health perception has a recall period of one year (Landgraf, Abetz, & Ware, 1999). Scores range from 0–100, where 100 indicates no impairment and zero indicates a severe impairment. The psychometric properties of the Swedish version have been reported by Norrby, Nordholm and Fast (2003) in a sample of 45 children with JIA. The reliability (Cronbach’s alfa) ranged between 0.86–0.94 and correlations to disease variables measured by the Child Health Assessment Questionnaire (CHAQ) were deemed strong, particularly in the physical domain (Norrby, Nordholm, & Fasth, 2003).

**Euroqol Five Dimensions Youth version (EQ5D-Y)**

This instrument is developed based on the standard adult version (EQ5D), which is a generic measure for health status that provides scores for different health dimensions and an index value for economic appraisal. The EQ5D-Y have the same five dimensions as the adult version, but with child-friendly wording, which was developed by a working-group. The instrument is developed for children between 7–12 years, but can be used up to the age of 15, at which point the adult version is recommended. Each
dimension has three levels of severity; no problems, some problems and a lot of problems with the recall period set at the same day as the response to the questions. The last item is a scoring of self-rated health on a vertical visual analogue scale ranging from 0–100 (“from the worst to the best health you can imagine”). The psychometric properties have been tested in a Swedish general population sample (n=407) and a high correlation to KIDSCREEN-27 was shown, with the kappa coefficient calculated to be up to 0.67 (Ravens-Sieberer et al., 2010).

The European KIDSCREEN-52
The KIDSCREEN project was funded by the European Commission during 2001–2004 in order to develop a measure for QOL and management of living resources for children aged 8–18 years. This instrument assesses children’s subjective health and well-being and is applicable for healthy as well as children who are ill. The instrument is cross-national and is developed in 13 European countries consisting of 52 items divided into ten different dimensions (physical well-being; psychological well-being; moods/emotions; self-perception; autonomy; parent relations/home life; social support/peers; school environment; social acceptance/bullying; and financial resources). The instrument items use a five-point Likert-type scale to assess either frequency or intensity with a recall period of one week (Robitail et al., 2006; The KIDSCREEN Group Europe, 2006). The psychometric properties have been evaluated by item response theory by using a cross-national sample (Sweden n= 3283). Internal consistency was measured using Cronbach’s alfa with a range between 0.77–0.89 for the subscales (Ravens-Sieberer et al., 2008).

Kid-KINDL-R
The purpose of Kid-KINDL-R is to measure HRQOL in healthy and unwell children and adolescents in the ages between 7–13 years. This instrument consists of 24+6 items with a recall period of the last week when answering to the questions. The first 24 items (generic) refers to six dimensions; physical well-being, emotional well-being, self-worth, well-being in the family, well-being related to friends/peers and school-related well-being. The last 6 items ask whether the child has any disease, and these items are referred to as a disease module. Thus, these items refer to a long-term condition in general. Response options are five categories and the scoring
is transformed to a 0–100 scale where higher scores indicates better HRQOL. The Kid-KINDL-R is validated with a large sample in Germany (n= 7649) and has been translated into 27 other languages. The psychometric properties are described as satisfactory with a Cronbach’s alfa at 0.63–0.76 for the German sample (Erhart, Ellert, Kurth, & Ravens-Sieberer, 2009).

**Paediatric Quality of life Inventory Generic core scales (PedsQL-4.0)**

This instrument is designed to measure the core of the health dimensions outlined by the WHO, and is exemplified by the dimensions of physical, emotional, social and school functioning. This instrument can be used both for healthy children and children with acute and long-term conditions. There are 23 items with a five-point ordinal scale to indicate how much the child describes problems with various areas of functioning. The recall period is one month and the instrument is transformed to a 100-point scale with lower scores indicating poorer HRQOL and clinical meaningful difference has been outlined (Varni, Burwinkle, & Lane, 2005; Varni, Burwinkle, Seid, & Skarr, 2003). Psychometric properties for the Swedish version (n=1570) have been tested by Petersen et al (2009), with results indicating Cronbach’s alfa value at 0.70 and a moderate result on test-retest (intra-rater reliability) (Petersen, Hägglöf, Stenlund, & Bergström, 2009).

**The DISABKIDS Chronic Generic Measure (DCGM-37) (-IV)**

The DISABKIDS questionnaire was part of the comparisons in paper I and was selected based on those results. DISABKIDS was used to measure HRQOL in paper II and was developed further in a web-based form which was used during the clinical intervention and a prerequisite for data collection to paper III–IV.

DISABKIDS has been developed in a European project initiated by the European Union (Sweden included) at the beginning of 2000. The intent was to develop and promote the use of standardized instruments for the assessment of HRQOL and to enhance the possibility to study HRQOL in a European perspective independently of the child’s type of long-term health condition. The development followed a bottom-up procedure, first with focus groups with children, in-depth interviews with some children...
and complementary focus groups with parents and professionals according to a standardized procedure. After item development and translations, several pilot studies were conducted in order to investigate psychometric analyses and field testing to assess retest reliability and construct validity in different contexts. DISABKIDS is a chronic generic measure, which deals with the HRQOL in children with different long-term conditions and aspects that pertain to a long-term condition in general. The self-report questionnaire is constructed for children aged 8–18 years. DISABKIDS consists of 37 Likert-scaled items assigned to six dimensions and a total score which is produced after a combination of the dimensions. Names of each dimension has been revised, since it has been a confusion of mixed positive and negative names of the dimensions (Chaplin et al., 2009). The new names of the dimensions are; “Independence” which is about the child’s future and life without impairment. The “Inner strength” is about emotional worries and “Social inclusion” is about the understanding of others. “Equality” describes stigma and whether the children feel that they are being left out. In the dimension of “Physical ability” the focus is on the functional limitations and perceived health status. The last dimension is “Treatment”, which involves descriptions of how the child considers the impact of taking medication (The DISABKIDS Group Europe, 2006). An overview of the dimensions and relation to WHO domains are described in Figure 3. For the DISABKIDS questionnaire, the respondents are asked to refer back to the last four weeks. Scoring is calculated from zero (poor HRQOL) to 100 (excellent HRQOL). The internal consistency on subscales have shown Cronbach’s alfa at 0.70–0.87 and test-retest reliability was deemed satisfactory with an intra-class correlation coefficient at 0.71–0.83 across different conditions (Simeoni, Schmidt, Muehlan, Debenasoson, & Bullinger, 2007).

In paper II, the disease-specific module for diabetes (DM-10) was used, which is complementary to the chronic generic module of DISABKIDS. The therapy of T1D exacts a serious impact on a child’s everyday life, such as planning daily routines, measuring blood glucose levels and injecting insulin several times a day. The QOL can be influenced by these procedures, but also because child’s fear of high blood glucose levels. The DISABKIDS Diabetes Module (DM-10) has two scales that measure the
“Diabetes Impact”, encompassing emotional reactions, to the feeling of needing control every day; and “Diabetes treatment” which refers to carrying equipment and planning treatment (Baars, Atherton, Koopman, Bullinger, & Power, 2005; The DISABKIDS Group Europe, 2006).

**Figure 3. The structure of DISABKIDS and each dimension and the relation to WHO domains (adapted and abbreviated) and the added disease-specific module for Diabetes**

**Development of web-DISABKIDS**

Before the start of the clinical intervention, a web-based form of DISABKIDS was developed in collaboration with the constructors and was built within the registry information technology system. The web-DISABKIDS has the same layout as the original questionnaire. A provision of information in each dimension in graphical displays in different colours (green - yellow - orange - red) and scores was produced by the algorithm of DISABKIDS. The green colours indicated good HRQOL, whereas red colours indicated worse HRQOL. Each colour represented cut-offs derived from the calculations based on the original algorithm, and was determined by the constructors of DISABKIDS. After answering, the child received a summary from the latest completion in a graphical presentation in colours on the screen (without scores) before signing out. In the decision support
system (DS), all results were displayed from all time points, with colours and with scorings of each dimension on the summary page. In addition, access to item level and each response option to the questionnaire was added as well as the ability to change the response option during the conversation to facilitate a user-friendly assessment tool. On the summary page, healthcare professionals could also observe other medical information, laboratory tests and treatments concerning the child’s clinical care.

The clinical intervention (Paper III–IV)

After web-DISABKIDS was introduced, a clinical intervention was performed. This was suitable for answering the question about how web-DISABKIDS could work in a clinical setting and to generate clinical knowledge regarding the use and assessment of HRQOL (Forbes, 2009). Information sheets about each dimension of web-DISABKIDS questionnaire and how to read and interpret the scores was constructed. During a 120-minute session at each clinic, the healthcare professionals received information about the registry system, HRQOL in general and how to use web-DISABKIDS in particular. This was performed by two instructors. Each clinic had prepared a separate place in the waiting-room with a computer or a tablet where the children could answer the questions. Before data collection, each clinic had used the web-DISABKIDS during a period of at least two months, in which the healthcare professionals had reviewed and discussed the results from the assessment tool with the child during consultations.

Measures from SWEDIABKIDS (Paper II)

The glycosylated haemoglobin (HbA1c) level is one important indicator when describing the care for children with T1D. The HbA1c target values indicates the level of metabolic control, which is linked to the long-term risk of complications, such as cardiovascular disease (Hanberger, Samuelsson, Lindblad, & Ludvigsson, 2008). HbA1c reflects the average blood glucose levels over the intervening three months, and higher values indicate poorer metabolic control (Rewers et al., 2007). The reference method recommended by the International Federation of Clinical
Chemistry (IFCC) has been adopted in Sweden, and the HbA1c values are referenced to as mmol/mol. The HbA1c target value has been determined to 51 mmol/mol due to guidelines from the American Diabetes Association, (American Diabetes Association, 2014). Consequently, the cut-off for HbA1c was set at 51 mmol/mol in paper II. Body mass index standard deviation (BMI-SDS) is a measure of the child’s relative weight, adjusted for age and gender. Given the age, gender and BMI of the child, an appropriate reference standard equivalent with the BMI for the age percentile could be determined (Karlberg, Luo, & Albertsson-Wikland, 2001).

HbA1c, BMI-SDS and disease duration were collected from SWEDIABKIDS, the Swedish paediatric diabetes quality registry (Registercentrum Västra Götaland, 2014). All laboratory methods used in Sweden are standardized through EQUALIS (External Quality Assurance in Laboratory Medicine in Sweden). The HbA1c data obtained from SWEDIABKIDS were derived from capillary blood samples measured with the Bayer/Siemens DCA-2000 analyser or by local laboratory methods (Shalitin & Phillip, 2012).

Semi-structured interviews (Paper III)

Semi-structured interviews based on open-ended questions were used to encourage children to talk freely about the topic of interest (Polit & Beck, 2013). The interviews were conducted individually, and were performed after the child’s regular appointment, in a separate room at the clinic. The purpose and type of event of interest may affect how easily children are able to communicate about the event. Children should be interviewed as soon as possible after they have experienced the event of interest, to warrant the most accurate information (Docherty & Sandelowski, 1999). It is beneficial to investigate how children interpret the questions and adapt questions to the individual child’s developmental stage (Kortesluoma, Hentinen, & Nikkonen, 2003). Therefore, an interview guide was developed prior to the interviews, and was pilot-tested with three participants. Changes were made in some of the questions after the pilot-testing, and was consequently not included in the sample. During the interview, attendant questions were asked to clarify what the children meant by asking “What do you think?” or “Could you explain more about
that?”. All interviews were conducted, recorded and transcribed verbatim by the author of this thesis.

Audio-video recordings (Paper IV)

The use of audio-video recordings to undertake naturalistic analysis of actions can provide opportunity to examine procedures and reasoning between participants. An understanding of ways in which technologies are deployed in consultations is possible when using this technique (Heath, Luff, & Sanchez Svensson, 2007). After consent from the healthcare professional as well as the child and parents, the consultations were audio-video recorded with a camera on a permanent tripod in the corner of the examination room. The focus was on the healthcare professional, and children and/or their parents were captured on the recordings if they were invited to join in front of the computer screen. In some cases, the recordings were capturing all individuals in the room, and in some cases, only the professional was captured. The researcher was not attending the consultations and the camera was operated by the professionals. Some recordings included the consultation from start to finish, and in some cases, only the conversations between the professional and child when the assessment tool was used. This was due to demands from the healthcare professionals who did not want to record the physical examinations during the consultation.

Data analysis

Quantitative analysis (Paper I–II)

In the first paper, a deductive, quantitative content analysis was chosen to perform an objective and systematic description of content which is governed by rules that are explicitly stated and applied (Krippendorff, 2004). The intent in paper I was to describe the content dimensions of HRQOL instruments from a biopsychosocial perspective and to the extent where the content in the instruments corresponds to and highlights the importance of experiences and activities that are relevant to children. Using the international classification of functioning, disability and health (version for children and youth; ICF-CY) is helpful to describe functioning,
activities and environments relevant to children. It has been shown that ICF-CY is applicable to obtain descriptions of child functional status, interventions and outcome measures (Adolfsson, Malmqvist, Pless, & Granlund, 2011; Björck-Åkesson et al., 2010; Ståhl, 2012). Beyond this biopsychosocial model, the classification can provide systematic coding schemes for health information systems (World Health Organization, 2007). When linking to the ICF a set of linking rules have been determined by Cieza et al (2005) (Cieza et al., 2005) which was used during the data analysis. The set of linking-rules are presented in Table 4.
Table 4. The eight specific linking rules for the linking process to ICF according to Cieza et al 2005 (adapted and abbreviated)

<table>
<thead>
<tr>
<th>Rule number</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Acquiring good knowledge of the ICF-CY framework (chapters, domains and categories)</td>
</tr>
<tr>
<td>2</td>
<td>Only linking each meaningful concept to the most precise category</td>
</tr>
<tr>
<td>3</td>
<td>Do not use the so-called “other specified” ICF categories</td>
</tr>
<tr>
<td>4</td>
<td>Do not use the “unspecified” ICF categories, instead use a lower level of category</td>
</tr>
<tr>
<td>5</td>
<td>If a meaningful concept is not sufficient, designation not definable (nd), should be used. Special cases in this rule: General health: nd-gh Physical health: nd-ph Mental health: nd-mh Quality of life: nd-qol</td>
</tr>
<tr>
<td>6</td>
<td>If a meaningful concept is clearly a personal factor defined by ICF-CY, this can be documented with pf for example age, gender, heredity or social background</td>
</tr>
<tr>
<td>7</td>
<td>If there is no evidence of a meaningful concept and no personal factors are identified, then an assignment of nc (not covered) is used</td>
</tr>
<tr>
<td>8</td>
<td>Health conditions or diagnosis should be assigned hc</td>
</tr>
</tbody>
</table>

The linking process followed several steps. After identification of the meaningful concept, the identification of components in the chapters of Body functions (b), Body structures (s), Activities and Participations (d) or Environmental factors (e) was determined. The second step was to find the chapter that the meaningful concept corresponded to, and the third step, selection was made of the appropriate level category for the meaningful concept. An example of the linking process is presented in Figure 4, where the meaningful concept “headache” has been linked to the ICF-CY.
In the second paper, descriptions were made using inferential statistics, and according to the categorical level and non-normally distributed data nonparametric tests were performed. The software program IBM Statistical Package for the Social Sciences Statistics 21.0 (SPSS) was used when conducting the analysis. Calculations of score were made by following instructions in the DISABKIDS manual and use of syntax files. Each scale was transformed to a range of 0–100 where higher values indicated better HRQOL (The DISABKIDS Group Europe, 2006). A p-value of <0.05 was considered statistically significant. Descriptive statistics (frequencies, percentages, means, ranges and standard deviations) were used to characterize data. A Mann-Whitney U test was performed to compare groups. Reliability as internal consistency was measured by using Cronbach’s alfa.
Qualitative analysis (Paper III–IV)

A qualitative content analysis was used to analyse interviews (Paper III) and video-recordings (Paper IV). A qualitative approach to content analysis is the close reading of texts or observations, which involves interpretations of texts or images into new narratives. The method is an attempt to emphasize differences and similarities from the categories derived during the analytical procedure when seeking the answer to the research question (Krippendorff, 2004). Content analysis makes it possible to test theoretical issues to enhance an understanding of the data, and if there is a lack of former knowledge about the studied area, or if the knowledge is fragmented, an inductive approach has been recommended (Elo & Kyngäs, 2008).

The qualitative content analysis with an inductive approach was applied and analysed in accordance with descriptions made by Elo and Kyngäs (2008), following different phases in the analysis. In the preparation phase, data units were selected to base the analysis on. This was the step to make sense of the data or to “become immersed with the data” which was made by reading through the texts several times and by watching the recordings several times. Meaning units were then sorted out which was based in the narrow descriptions of the data. The next phase, called the organising phase, was the process in which the open coding and creating of categories began. Notes and headings were written down while reading the text and while watching the recordings, to describe all aspects of the content related to the study aim. The notes from the readings were encoded to coding sheets by using Microsoft Excel (2010). The codes were sorted into subcategories and reduced by collapsing those that were similar into a higher order category by a classification of their attributes which implied a comparison between the codes and this was made by using different colours on the codes. Finally, the subcategories were abstracted into generic categories which described the essence and underlying meaning of the subcategories (Elo & Kyngäs, 2008).

For this thesis’ specific aims, a secondary analysis was performed. In the analysis of results from paper II, a deductive approach of the results was used based on ICF-CY, to illustrate a biopsychosocial perspective. The analysis followed the same procedure which have been described earlier.
(paper I). Also, a comparative analysis between results from paper III and IV was conducted. The analytical approach used was a concept analysis, which is a process of clarifying and describing relations between concepts (Nuopponen, 2010).

Ethical considerations

Regardless of methodology, the protection of the child’s interest as well as risks or benefit calculations have to be done regularly during the research process. Also, the UNCRC addresses respect for dignity and worth for each child, and respect for the views of the child (UNCRC, 1989). In accordance with Swedish law, the studies in this thesis were approved by The Regional Ethical Review Board at Linköping University, Sweden (Dnr 2012/249-31 and Dnr 2013/107-31). There are three basic ethical principles which need to be considered; autonomy, beneficence and justice.

Autonomy

The respect for each individual incorporates the respect for autonomy as well as the protection of persons with impaired or diminished autonomy (Councils for International Organizations of Medical Sciences CIOMS, 2002). In this thesis, informed consent with verbal and written information to the participants is considered as a way to cover for autonomy. An informed consent can be withdrawn without prejudice at any moment. The consent requires the person to be competent and children (0–15 years) are —due to their developmental and cognitive limitations—not yet capable of providing legal consent to participate in research (Alderson & Morrow, 2011). Therefore, the information about participation must be adapted for children at their cognitive level. Parents/legal guardians have to give their informed consent (Altavilla, 2011) to children 0–15 years of age, and children must provide their assent. Age-appropriate information must be provided to enable the child to make well-considered decisions about taking part in a research project or verbalize their right to withdraw (Matutina, 2009). The information letters were age-adjusted with different wordings for children between the age of 8–12 and 13–18. Written information was given before data-collection, in
which a nurse at each department had sent the information letter by mail to the family. The researcher provided verbal information to the participants when meeting them at the clinic, before data-collection took place.

**Beneficence**

Beneficence refers to ethical obligations to maximise benefits and to minimise risk to harm the person (Councils for International Organizations of Medical Sciences CIOMS, 2002). The ethical principle of respect requires children to be involved in decision-making if it is possible when the child’s age and maturity is taken into consideration. However, it is questionable whether research risks can be justified when children themselves have no interest of their own in a research project. Children and their families have to face the difficulty in making decisions based on contingent information (Pinxten, Nys, & Dierickx, 2010). The researcher should be sensitive to signs that may reflect wishes from participants to end their participation. Additional verbal clarifications were given about the video recordings, in which the focus was on the healthcare professionals, and not on the child or parent. This was important to clarify as there is a risk of experiencing encroachment of integrity when being video-recorded. The participants were informed about confidentiality, which was obtained by securely storage of the video-recordings, as well as for the taped and transcribed interviews. In the reporting of the data, it was ensured that none of the participants could be identified.

**Justice**

The third principle is about justice, which refers to the obligation to treat each person properly (Councils for International Organizations of Medical Sciences CIOMS, 2002). Paediatric research should offer child-friendly settings and procedures that limit the invasiveness to a minimum (Altavilla, 2011). The researcher should use methods that give children opportunity to provide their views and researchers have to be responsive to each child’s own agenda (Kirk, 2007). The maturity and awareness that children possess should not be underestimated. When children are given
the opportunity to express their views they can gain confidence. If any risk is suspected concerning the child, researchers have to give the appropriate response and take responsibility to act in the best interest of the child, which would guarantee that children’s voices will be heard in research (Miller, 2000). A limitation and a threat against the principle of justice is that children who cannot speak and understand Swedish have been excluded in this project. The inclusion criterion was based on the questionnaire restrictions. Although the participants gain little or nothing by participating in this research, the overall research may be justified when seen in a wider perspective.
Results

The results are presented on the basis of the two specific thesis aims, which are built on the four papers.

HRQOL among children with T1D according to a biopsychosocial perspective

The content dimensions of HRQOL instruments according to ICF-CY

The instruments included 235 items altogether, which resulted in the identification of 290 meaningful concepts, of which 258 corresponded to codes within the ICF-CY, and accounted for 89 % coverage. Concepts not linked to ICF-CY (11 %) dealt with general health (n 24), QOL (n 1) health conditions (n 3) and concepts not covered (n 4) which were named not covered. The representations of ICF-CY components are described for each instrument in Table 5.

The overall representations of body functions in the instruments accounted for 31 % of the total, which was described by temperament and personality functions, for example optimism and psychic stability. Other aspects of body functions that are encompassed by the classification are body image and experience of self, pain and sleep functions. The representations of activities and participation had the largest part within the instruments with 46 % coverage. This classification describes general tasks and demands, such as undertaking multiple tasks and handling stress or other psychological demands. Other aspects that are covered include mobility and self-care which is about moving or changing body positions and caring for oneself, for example dressing, eating and washing. Basic interpersonal interactions which is about interactions with other people are covered by all instruments. School education and engagement in play, shared cooperative play, recreation and leisure such as sports and
socializing was also identified. *Environmental factors* accounted for the smallest proportion, with 12% coverage. This classification covers influences such as the use of drugs and financial assets, support from healthcare professionals, relationships from family—for example of immediate family members—and friends. Additionally, the influences of the individual attitudes of acquaintances, peers, colleagues, neighbours and community members was exemplified.

**Table 5. Representations of ICF-CY components in the instruments described by number and percent from the total number of 258 meaningful concepts identified**

<table>
<thead>
<tr>
<th>Instrument</th>
<th>Body Functions</th>
<th>Activities and participation</th>
<th>Environmental factors</th>
<th>Other: QOL, gh, hc, nc*</th>
</tr>
</thead>
<tbody>
<tr>
<td>CHQ-CF</td>
<td>29 (27%)</td>
<td>58 (55%)</td>
<td>5 (5%)</td>
<td>14 (13%)</td>
</tr>
<tr>
<td>DCGM-37</td>
<td>15 (29%)</td>
<td>24 (47%)</td>
<td>9 (18%)</td>
<td>3 (6%)</td>
</tr>
<tr>
<td>EQ5D-Y</td>
<td>4 (24%)</td>
<td>11 (65%)</td>
<td>0</td>
<td>2 (11%)</td>
</tr>
<tr>
<td>KIDSCREEN-52</td>
<td>18 (33%)</td>
<td>17 (31%)</td>
<td>13 (24%)</td>
<td>7 (12%)</td>
</tr>
<tr>
<td>Kid-KINDL-R</td>
<td>15 (44%)</td>
<td>8 (24%)</td>
<td>6 (18%)</td>
<td>5 (14%)</td>
</tr>
<tr>
<td>PedSQL 4.0</td>
<td>9 (33%)</td>
<td>14 (52%)</td>
<td>3 (11%)</td>
<td>1 (4%)</td>
</tr>
</tbody>
</table>

*Meaningful concepts linked to Quality of Life, General Health, Health Condition and Not Covered

According to the ICF-CY framework, a considerable focus of the results were on *Activities and participation* from a biopsychosocial point of view, with *Body functions* also being relatively well covered. The least significant part was on *Environmental factors*. When each instrument was reviewed, the DCGM-37 and KIDSCREEN-52 were comparable between ICF-CY components (Table 5). Due to these results, DISABKIDS was selected and the disease-specific module for diabetes (DM-10) was added to describe the HRQOL of children with T1D. The DM-module contained 17 meaningful concepts which was linked to the ICF-CY, with 6 codes linked to *Body functions* (35%), 4 codes linked to *Activities and participation* (24%) and
4 codes linked to Environmental factors (24%) respectively. Only three additional codes were used; two codes denoted to health condition and one denoted to not covered.

**Differences in HRQOL of children with T1D**

The sample of children with T1D was compared in groups, by dividing them by age ranges (8–12 and 13–18) with boys and girls and according to metabolic control (HbA1c <51 mmol/mol versus HbA1c >52 mmol/mol). The results showed that the mean in all dimensions of DISABKIDS was decreasing for adolescents (13–18), for girls and for children with higher HbA1c (Figure 5).

![HRQOL presented in each dimension of DISABKIDS (children with T1D)](image)

**Figure 5. Mean in each group according to DISABKIDS dimensions.**

The dimension with statistical significance across all groups were the dimension of Inner strength. “Inner Strength” corresponded to ICF-CY codes in both body functions and activities and participation. The items
within this dimension were coded to emotional functions and temperament and personal functions, and psychological stability. The codes from activities and participation covered the ability to handle stress and other psychological demands, as well as acting predictably and maintaining one’s own health (Table 6). In the dimension Treatment, the statistically significant difference between genders and HbA1c were found. Items in this dimension corresponded to looking after one’s health, for example about well-being and handling stress, which is found in the chapters of Activities and participation. In the ICF-CY chapters about Body functions, items in this dimension described emotional functions and range of emotions, psychological stability and memory functions in relation to drugs (Table 6). There were also differences between genders and metabolic control in the dimensions of the DM module. Items in Diabetes Impact correspond to thought functions, attention functions and emotional functions in the chapters about Body functions. The code d570 about looking after one’s health was also described here. Environmental factors were covered by the codes about products or substances for personal consumptions and products and technology for personal use in daily living. Items in Diabetes treatment was related to the same codes found in the dimension of diabetes impact (Table 6).
Table 6. Description of DISABKIDS and DM-10 linkage to the ICF-CY and statistical significant difference between groups in the sample from children with T1D (p<.05)

<table>
<thead>
<tr>
<th>Dimension in DISABKIDS (item no.)</th>
<th>Items linkage to ICF-CY codes</th>
<th>Girls and boys</th>
<th>Age groups</th>
<th>Metabolic control</th>
</tr>
</thead>
<tbody>
<tr>
<td>Independence (1–6)</td>
<td>b1265, d570, d220, b125, d2202, b110, nd qol</td>
<td>ns</td>
<td>ns</td>
<td>.023</td>
</tr>
<tr>
<td>Inner strength (13–19)</td>
<td>d5702, b152, b1263, b152, d240, b126, d2503</td>
<td>.006</td>
<td>.010</td>
<td>.005</td>
</tr>
<tr>
<td>Social inclusion (26–31)</td>
<td>d7102, d9103, d8803, d920, d155, d7101, d330</td>
<td>.023</td>
<td>ns</td>
<td>ns</td>
</tr>
<tr>
<td>Equality (20–25)</td>
<td>d710, e455, b1400, d820, e425, e,425, b1800</td>
<td>.020</td>
<td>ns</td>
<td>ns</td>
</tr>
<tr>
<td>Physical ability (7–12)</td>
<td>d4552, d4551, b1300, b1252, d330, d720, b134, d880, d920, nd gh</td>
<td>ns</td>
<td>ns</td>
<td>.021</td>
</tr>
<tr>
<td>Treatment (32–37)</td>
<td>d240, b144, b1263, b152, b1522, d570, e1101x6</td>
<td>.004</td>
<td>ns</td>
<td>.017</td>
</tr>
<tr>
<td>Diabetes Impact (3–8)</td>
<td>b140, b152, b510, d550, d570, e110, e115, nc</td>
<td>.023</td>
<td>ns</td>
<td>.007</td>
</tr>
<tr>
<td>Diabetes treatment (1–2, 9–10)</td>
<td>b140, b160, d570, e115, hc</td>
<td>.011</td>
<td>ns</td>
<td>.029</td>
</tr>
</tbody>
</table>

ns: not statistically significant, age-groups 8–12 and 13–18, cut-off for normal HbA1c< 51 mmol/mol
Children’s and healthcare professionals’ use of a HRQOL instrument

Children’s experiences about a structured assessment of HRQOL

Children described seven aspects after they had answered the DISABKIDS questionnaire and discussed their results with their professional in the encounter. The assessment was confirming their health by giving them a summation which was easy to understand and offered a picture about how they were feeling. An opportunity to improve their health was possible when the dimensions of HRQOL was displayed to them in the coloured fields. After answering, children could understand more about which aspects needed more attention. This was also helping the children to understand and describe their health, which was interpreted as increasing their knowledge about health, as results were presented in a way that was easy to interpret and made sense to them. When looking at the results, they could be discovering variations over time if they had answered to web-DISABKIDS before, since it was easy to visually detect similarities and differences based on the coloured fields. Children described that by sharing with professionals, they could get a general picture of their health in addition to medical issues, which was important to discuss with the healthcare professional, preferably without parents present. By discussing with professionals, children obtained cause-related explanations which could give them an understanding of how the long-term condition influenced and affected everyday life. Reasons could sometimes be explained by aspects in their everyday life (such as sleeping overnight with friends). By receiving feedback, children could be offered support from professionals which motivated children to answer to the questionnaire, and it was experienced as only sufficient to answer if healthcare professionals were reviewing the HRQOL results during the encounter.
How healthcare professionals use a structured assessment tool for HRQOL

During patient encounters, healthcare professionals used results from the assessment in various ways, leading to different approaches. Each approach consists of combinations of different overlapping strategies that professionals were using during conversations (Table 7). The first strategy; “describes the assessment tool and asks for understanding” was used in all three approaches. In the instructing approach, professionals used the visual image to capture an overview of the child’s situation and they gave children a short summary of the findings of the HRQOL results as it was presented on the summary page. When giving explanations of results, professionals related to the child’s long-term condition. In the inviting approach, children were invited to select dimensions to talk about, and healthcare professionals were awaiting the child’s response. When discussing causes, professionals aimed to connect to aspects in child’s everyday life. Moreover, professionals used the visual image as a starting point for further discussions and asked the child how they experienced their situation and then connected to results from the assessment tool. Healthcare professionals were seeking motivation for change and discussed problem-solving with the children. By an engaging approach, professionals explored each dimension based on the underlying items in the assessment tool and sometimes changed their response option after discussions with the child. At the end of the conversation, they identified common aspects to further work on and discussed these aspects together with the children. When an engaging approach was used, professionals were seeking a common interpretation of results together with the child at the end of the encounter.
Table 7. Description of each strategy used by the professional and how the strategies overlap each approach

<table>
<thead>
<tr>
<th>Strategies</th>
<th>Instructing approach</th>
<th>Inviting approach</th>
<th>Engaging approach</th>
</tr>
</thead>
<tbody>
<tr>
<td>Describing the assessment tool and asking for understanding</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using the visual image to capture an overview of the child’s situation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using the visual image as a starting point for further discussion</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Asking how the child experiences the situation and then connects to the assessment tool</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Exploring each dimension based on the underlying items</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Seeking a common interpretation of the results from the assessment tool</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Comparison between children’s experiences and professionals’ approaches

The results from children’s experiences and healthcare professionals’ use of the assessment in the encounter was compared using a concept analysis. The comparison is based on all seven aspects described by children and the three approaches that explored how healthcare professionals were using the assessment tool. The comparison showed that when healthcare professionals were using an instructive approach, only three aspects were covered as described by the children (Table 8). Children could be offered a confirmation of their health when professionals gave a summary of the results from the basis of the summary page. Healthcare professionals gave explanations about causes, which was appreciated by the children. The case in the instructing approach was a simple use of the assessment tool, which was revealed when comparing to the children’s experiences. When the inviting approach and engaging approach was used, all seven aspects described by children were addressed. This was understood as promoting
the child’s involvement, since children described the importance of sharing their results with the professionals and receiving feedback which implied support to them. According to the comparison, there were no differences between the inviting approach and the engaging approach as revealed from the children’s statements. This could be explained by the fact that each approach had strategies that were overlapping (Table 7). The comparison between children’s descriptions and how professionals were using the assessment during the encounter is visualised in Table 8.

Table 8. Comparison between children’s experiences and healthcare professionals’ use of the HRQOL assessment tool in encounters

<table>
<thead>
<tr>
<th></th>
<th>Instructing approach</th>
<th>Inviting approach</th>
<th>Engaging approach</th>
</tr>
</thead>
<tbody>
<tr>
<td>Confirming health</td>
<td>x</td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Opportunity for improving health</td>
<td>x</td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Increasing knowledge about health</td>
<td></td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Discovering variation over time</td>
<td>x</td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Sharing with professionals</td>
<td>x</td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Obtaining cause-related explanations</td>
<td>x</td>
<td>x</td>
<td>x</td>
</tr>
<tr>
<td>Receiving feedback imply support</td>
<td></td>
<td>x</td>
<td>x</td>
</tr>
</tbody>
</table>
Discussion

Methodological considerations

Throughout the studies in this thesis there were different ways to ensure trustworthiness of the research findings. Since papers I and II have a quantitative approach, validity and reliability concepts are used. A naturalistic inquiry was used in papers III and IV and therefore aspects concerning qualitative perspectives are considered.

*Trustworthiness in quantitative studies*

The four concepts *internal validity*, *external validity*, *construct validity* and *statistical conclusion validity* is used and each type is pivotal as they convey several of the considerations (Kazdin, 2003). Also, reliability issues are used to describe the methodological considerations of paper I and II.

Internal validity

Internal validity refers to what extent the study makes plausible explanations of the results. If any variable outside of the study occurs, that must be accounted for in the results (Kazdin, 2003). To achieve internal validity, confounding factors should be reduced to a minimum. It is worth mentioning, that there is almost never a completely determined cause assignable to any given study. The data collection in paper II had limitations on account of the clinics that did not register how many children they had asked for participation. Only the number of participants that responded positively was registered, and thereby the drop-out rates were calculated on that basis. The attrition analysis showed that non-responders had a higher HbA1c than responders, which is a threat against internal validity and is a potential risk for systematic errors (Brossart, Clay, & Willson, 2002). Whether children had help from parents when answering is not known, which could be another threat against the internal validity of the results from paper II.
External validity

The external validity refers to the question about the boundary conditions of the findings (Kazdin, 2003), often discussed as generalization or applicability of the findings. One key to external validity is a rich description of the population and contextual information (Glasgow et al., 2006), which could support practitioners to better judge the applicability of the results to other settings in practice. A threat against external validity is the question if the same results would occur if the participants were not aware of being part of a research study (Kazdin, 2003), which was not possible in the studies of this thesis, due to ethical issues. An attempt to strengthen the external validity was by having few exclusion criterions in paper II, however, it could be argued that the sample size was relatively small which could be a threat against the external validity. Nevertheless, since the results had been revealed in other research (Chaplin et al., 2009; Hanberger, Ludvigsson, & Nordfeldt, 2009), this could be viewed as strengthening our results. The description of the operationalized concepts of the instruments in paper I was an important step to strengthen the external validity, however, there are shortcomings regarding the descriptions of each instrument in what they are measuring, which must be seen as a limitation and a threat to the external validity concerning the results in paper I.

Construct validity

The conceptual basis for the underlying effect is known as construct validity (Kazdin, 2003). The description of the concept of HRQOL needs to be defined and dimensions measured, and a theoretical reasoning for the sample of the specific items is discussed in paper II. A factor analysis can investigate the construct validity, which has been performed by the constructors of the questionnaire. The construct of HRQOL is well described and DISABKIDS has shown sufficient psychometric properties (Simeoni et al., 2007). Using the ICF framework to describe the content in instruments, may lead to a loss of information about the context and focus of the items. On the other hand, mapping the constructs to ICF could serve as a guide when selecting an appropriate instrument (paper I). The point is to pay attention to whether there is a particular functioning or well-
being that is being measured (Janssens et al., 2015), which was confused in the compared instruments (paper I).

Statistical conclusion validity

The studied population was a group of children with the same condition (T1D), which could strengthen the statistical conclusion validity. A threat against this type of validity is the lack of power analysis or effect size (Kazdin, 2003) which was not estimated. Also, type I and type II errors need to be addressed. Selection bias is one aspect that could have affected the results, since participants that chose to answer a questionnaire about HRQOL could be more willing or able to answer that kind of questions, or could felt pressured to answer, which is potential biases (Boynton, 2004). Thus, there are several criteria for the judgement of causal associations (Grimes & Schulz, 2002), and in terms of paper II, the consistency of associations were found in other studies measured with the same instrument (Hanberger et al., 2009; Petersson, Huus, Samuelsson, Hanberger, & Akesson, 2015). Also, biological plausibility was assured as the associations made sense in the context of a clinical practice.

Reliability issues

Methods for assessing reproducibility—also known as reliability—include agreement between different observers who are using the same technique (Kirkwood & Sterne, 2003). In paper I the inter-rater reliability was tested in a specified procedure. The first step involved two researchers in the linking process. In step two, every tenth item was selected from each instrument and an independent researcher coded these items with the same procedure and rules that had been specified. The inter-rater reliability was 74 %, which was deemed acceptable according to other studies using the same coding process (Adolfsson et al., 2011; Ståhl, 2012). Discussion about differences was made in between coders to reach agreement, which strengthen the reliability of the coding procedure. In paper II, the internal consistency was measured with Cronbach’s alfa. If items within a scale intends to measure aspects of the same construct, they should be correlated to each other. A common rule is that a scale should have a minimum alfa value of 0.7 (Kirkwood & Sterne, 2003). The results
from DISABKIDS generic scale and the diabetes specific scale showed satisfactory internal consistency with an alfa value of 0.94 for the generic part and of 0.88 for the diabetes module.

**Trustworthiness in qualitative studies**

The constructs of credibility, dependability, confirmability and transferability, are frameworks for ensuring rigor in qualitative research (Lincoln & Guba, 1985) and are used to describe the methodological considerations of paper III and IV.

**Credibility**

There are several ways to establish credibility, which deals with the confidence in how well data is addressing the intended focus (Lincoln & Guba, 1985). One example to establish credibility is by using pilot interviews to determine suitable interview questions to obtain rich data that provide answers to the proposed research question (Elo et al., 2014), which was the case in paper III. After pilot interviews, the interview guide was improved, and therefore the pilot-interviews were not included in the sample. The use of member-checking and peer debriefing by another experts who reviews and scrutinizes the findings is another way to demonstrate the credibility (Morse, 2015). Peer-debriefing was applied by discussion of results in a group-seminar (paper III and IV). In paper III, the participants had different kinds of long-term conditions and a variety in how often they attended the clinic. There were more girls than boys which could be viewed as a limitation. In paper IV, member-checking was used despite this method has been criticized. Two participants were introduced to the strategies (subcategory level) and they confirmed these strategies and thereby strengthened the credibility. The range of video-recordings for each participant varied to between one and six recordings. The number of recordings could be regarded as a strength when the professionals changed their ways with respect to using the assessment tool dependent on the specific situation.
Dependability

Dependability refers to the replicability and consistency (Lincoln & Guba, 1985). Therefore, the descriptions of inclusion criterion is crucial as well as to further address the processes how the study has been conducted by reporting details to increase the ability to repeat the work (Shenton, 2004). The sampling procedure was based on a convenient sampling. Considerations about sampling are whether there is an appropriate sample of participants that best represents, or has the knowledge about the topic of interest. Therefore, departments were selected based on their previous experience of using the quality registry in clinical practice. This could have influenced the dependability, since departments included had a positive attitude to use the registry in general. Data was reviewed and coded by an analyst with experience in coding qualitative data and was supervised by the research team members. Discrepancies between codes were examined further and reviewed until consensus was achieved to establish the dependability of the results in paper III and IV.

Confirmability

Potential coder bias was extenuated through periodic “check-ins” on the coding process by several of the researchers as well as by relating the codes to the data. Another way to establish confirmability is by audit trail: providing details of the data analysis and which decisions that lead to the findings (Lincoln & Guba, 1985; Wolf, 2003), which were used in paper III and IV. The analytic procedure has been carefully described to facilitate the possibility to repeat the analysis. The analysis has also been following descriptions made by Elo and Kyngäs (2008) which also strengthen the confirmability. Recommendations were followed in which one researcher was responsible for the analysis and the other members of the research team followed up the analysis by discussions of divergent options about categorizations (Elo et al., 2014).

Transferability

A way to establish transferability is to use thick descriptions (Lincoln & Guba, 1985) by describing the participants and settings. Moreover,
demonstrating the research rigor by describing the procedure and how categories were emerged. The intent in qualitative inquiry is firstly not to generalize the results to other settings, it is rather to increase the understanding of the context or particular characteristics of the organization in which the fieldwork was carried out (Shenton, 2004). This research is limited to the context where the quality registry system is used, which is mainly a Swedish phenomenon, and limits the transferability to other settings. On the other hand, the questionnaire is used in other research settings which in turn could provide the ability to transfer the knowledge generated from this research outside the Swedish context.

Methodological considerations according to specific thesis aims

A concept analysis was used to compare the results from papers III and IV. This analysis was chosen in order to synthesize results and draw conclusions (Nuopponen, 2010). The concepts were derived from results based on former content analysis, which had taken processes for trustworthiness under consideration. A critique to concept analysis is the lack of consensus about how to perform such analyses. However, an emphasis on clarifying relations between concepts and how to perform such an analysis in detail have been described by Nuopponen (2010), which was used to guide the comparison of concepts derived from papers III and IV.

The use of a clinical intervention

A clinical intervention was used to introduce the assessment tool of DISABKIDS at the participating clinics. It could be argued that the standardization of the clinical intervention was lacking since it was not following a protocol and neither assessments were used to investigate how the clinical intervention worked (Forbes, 2009). However, the effect of the clinical intervention was not the purpose in this research, rather to describe and explore how the assessment tool was experienced by the children and could be used by healthcare professionals in a clinical practice.
Discussion of the results

This thesis comprises studies which explore and describe the use of instruments about health-related quality of life in outpatient care for children with long-term conditions. The results show that based on a biopsychosocial perspective, generic and disease-specific instruments can be used to gain a comprehensive description of the child’s HRQOL. On the basis of the results in paper I the DISABKIDS instrument was selected, which is applicable when describing different aspects of health concerning children with T1D. Children expressed positive experiences when they answered to the DISABKIDS questionnaire prior to their consultation, but also described the importance of sharing their results with the healthcare professional. When healthcare professionals are using DISABKIDS as an assessment tool in the encounter, three different approaches emerged, leading to different outcomes for each child. When an instructing approach is used, the child’s ability to learn more about their health and how to improve their health is limited. When an inviting or engaging approach is used by the professional, the child may become more involved during the conversations.

Understanding differences in HRQOL

It could be concluded that the knowledge about differences in HRQOL between different groups, and specifically which dimensions that are being affected, may be helpful when target interventions are planned. Healthcare professionals can use HRQOL instruments as a complement to biomedical variables to receive a biopsychosocial perspective on the child’s health.

HRQOL in relation to child’s age and gender

In this thesis, gender and age differences are found in children with T1D, with girls reporting lower HRQOL than boys. Differences between boys and girls is well known: girls report worse HRQOL than boys because of worries about the disease (Al-Akour, Khader, & Shatnawi, 2010). This indicates the vulnerability of teenage girls (Hanberger et al., 2009), which
is important knowledge for healthcare professionals who meet these children at the clinic. It has been found that nurses generally support person-centredness in their care through nursing activities, however, nurses should take the personal life of each person into account in a way that supports individuality in the care regularly (Suhonen et al., 2011). This should be included for all professionals involved in the care of children with long-term conditions.

Results show that HRQOL tends to decrease when children are entering adolescence, specifically concerning emotional functions and psychological demands. This implies a challenge for the healthcare infrastructure, to mitigate any potential long-term negative effects on an adolescent’s perceived HRQOL (Varni, Limbers, & Burwinkle, 2007). It could be argued that the perceived adolescent life satisfaction can be related to poor physical health (Zullig, Valois, Huebner, & Drane, 2005). As a result of the physical development during puberty, adolescents may feel impaired sense of well-being, which can explain the decreasing HRQOL during adolescence (Bisegger, Cloetta, von Bisegger, Abel, & Ravens-Sieberer, 2005). However, it has been argued that adolescents living with a long-term condition generally describe their health as the same way as their healthy peers (Taylor, Gibson, & Franck, 2008), which highlights the need to understand the personal perspective for each child. Furthermore, care should be planned not only based on clinical routines and predetermined guidelines for the specific condition, but instead needs to be co-produced in agreement with the child and the family. This could be supported by Engels biopsychosocial model, which proposes a shift from understanding disease solely as a physiologic condition to also include the persons’ own experience (Borrell-Carrió, Suchman, & Epstein, 2004).

**HRQOL in relation to metabolic control**

Results revealed a lower HRQOL in physical and mental domains of those children with poor metabolic control, but not in the social domains of DISABKIDS. This implies that the care of children with T1D should be focusing on improving metabolic control as well as implementing the screening of depressive symptoms. Unarguably, there is clinical relevance
for screening for depression among adolescents (Corathers et al., 2013), and it has been suggested that aspects concerning mental health should be included in the initial and in follow-up assessments of children with long-term conditions (McDougall, Wright, DeWit, & Miller, 2014). It is also worth mentioning that it is important to look beyond subgroups within disease categories, since there is a variety depending on the disease severity (DeWalt et al., 2015). Monitoring metabolic control is important because HbA1c levels during the first year may predict the level of metabolic control in the following years after diagnosis (Åkesson, Hanberger, & Samuelsson, 2015). The paediatric diabetes team should be aware of the impact on good metabolic control during adolescence, because it is important to reduce risk for complications in young adults. By working with adolescents and focusing on the vulnerable times in life, this could prevent the subsequent effects of poor metabolic control (Anderzén, Samuelsson, Gudbjörnsdottir, Hanberger, & Åkesson, 2016), as well as helping adolescents to better cope with their feelings of worries and fears about the future. HbA1c presents average glucose values over time and if used without describing the patterns that are essential to make informed decisions about treatment adjustments, the HbA1c value is limited (Corathers et al., 2015). Therefore, technologies that support the information visualization of data sharing about metabolic control as well as HRQOL are essential.

The results also indicate that items about treatment and disease-specific items about impact of T1D are reported worse by children with high HbA1c, which is described in terms of limited ability to handle stress and emotional functions. It has been stated that children need support in clinical practice to better cope with and manage the treatments of T1D, which has potential to improve children’s overall HRQOL (Naughton et al., 2014). This is another argument for the clinical applicability of HRQOL instruments in the management of care for children with long-term conditions.
Biopsychosocial perspective on using HRQOL instruments

It could be concluded that generic and disease-specific questions may gain a biopsychosocial perspective on health and illness. The distribution of ICF-CY components in the compared instruments has its main focus on activities and participation (paper I) which also have been found by others (Gandhi et al., 2014; Riva, Bullinger, Amann, & von Mackensen, 2010). Although, in the disease-specific module for T1D, more items were related to environmental factors. ICF-CY as a framework gives a valuable opportunity to capture factors that are influencing physical and emotional functioning, activities and participation in instruments about HRQOL (Gandhi et al., 2014). Nevertheless, some limitations of ICF need to be addressed. For instance, emotional functions which are explained by emotions such as sadness, fear or laughter (b152) do not distinguish between the differences in these concepts. This is because the components of ICF are focused primarily on objective functioning rather than the subjective (being sad or being happy) (Gandhi et al., 2014). Planning and evaluating care for children with long-term conditions can be grounded in the framework of the ICF but the classification needs to be improved to consider these differentiations.

When results from HRQOL instruments are used during consultations, children may gain an understanding of their health and illness. Children describe that by receiving their results from the assessment of HRQOL, they were provided with insights about their health which constitute a motivation for change. This implies that assessments of HRQOL generates information that can provide a basis for improvements in the care for children with long-term conditions (Norrby, Nordholm, Andersson-Gäre, & Fasth, 2006). Furthermore, the use of questionnaires can reveal additional information about the child’s psychosocial functioning (Boogerd et al., 2015). Certainly, all kinds of information from decision support systems and self-management plans can be effective, when they are used as a supplement rather than replacing the interaction between the healthcare professional and the child (Coulter & Ellins, 2007). It is important to use open-ended questions that pay attention to the child’s own concerns and allow time for the child to express themselves.
(Robinson, Callister, Berry, & Dearing, 2008). This could be a challenge when using standardized questionnaires in clinical practice, if the questionnaire is used to dominate the conversations and not used as a support.

The use of instruments for measuring HRQOL has limitations and the discrepancy between the assessment of children’s functioning (what the child can do) and well-being (feels about the situation) needs to be addressed. Several of the HRQOL instruments seem to confuse these concepts (paper I), which also have been found by others (Janssens et al., 2015; Solans et al., 2008). HRQOL instruments can measure health-related domains by levels of functioning that addresses biological, psychological and social components; however, what distinguishes the functioning is how the domains are measured. One example is absence, frequency, capacity or severity, in comparison to HRQOL measures which often have their focus on expectations, standards or concerns about the health domain (Fayed et al., 2012). Another aspect is whether the instrument is suitable for detecting change, or if the intended use is to describe functioning or perceived health status in a certain situation (Coster & Khetani, 2008). There is still little agreement in the literature about what generic HRQOL instruments intend to measure, which highlights the need for conceptual distinctions about HRQOL and biopsychosocial aspects of health (Fayed et al., 2012). Concurrently, from a system perspective, health and functioning cannot be separated from other aspects of life. ICF is a biopsychosocial model which encompass that all domains that comprise life also have an impact on the child’s QOL (McDougall, Wright, & Rosenbaum, 2010). When assessing a child’s health and functioning, aspects concerning QOL should also be considered to obtain a complete picture. It is suggested that QOL is described in ICF-CY, as a composite, emergent and changing phenomenon of different aspects of functioning that may have an impact on the child’s health condition, environmental factors and personal factors (McDougall et al., 2010). To conclude, both functional capacity and performance, environmental and personal factors are related to the perceived QOL, which indicates the benefits for assessing and enhancing QOL in children with long-term conditions (McDougall et al., 2014).
Sharing HRQOL results

Results in this thesis revealed that children expressed that it was encouraging if outcomes were requested and discussed together with the healthcare professional. This result suggests that it is important to identify the unique factors that influence the everyday life of children with long-term conditions (McDougall et al., 2010). Children have described that being in command and managing their life by themselves is important (Huus & Enskär, 2007). Concurrently, a good communication with the healthcare professional may result in a positive experience if children are encouraged to join in decision-making. Children can also gain by the learning opportunities arising in the encounter, which may promote their confidence in managing their disease (Lowes et al., 2015). It could be emphasized that the child’s knowledge and understanding can be improved and children may benefit when they are involved in their care (Coulter & Ellins, 2007), which was expressed by the children. Healthcare professionals should view each child’s results of HRQOL in the encounter, because HRQOL is subjective and dynamic and unique to each person, and includes different aspects of physical, psychological and social functioning (Taylor et al., 2008). It could also be argued that an increased use of a biopsychosocial view on health has implications for all health professionals (Hwu, Coates, & Boore, 2001). This implies that working in partnership with children and their family, possibly over extended periods of time are needed. It has been found that children are aware of many implications and useful strategies for managing their disease. However, if children are not included in the setting of goals and creating of the strategies to meet these goals, they may not develop the appropriate skills for participation in the management of treatment and self-care, which is a prerequisite when entering young adulthood (Beacham & Deatrick, 2015). This points to the conclusion that children need to receive knowledge and skills to achieve their personal goals concerning their health and effects on everyday life.

Children expressed that it was insufficient to answer to the questionnaire prior to the visit if the results were not reviewed and discussed by the healthcare professional. This highlights that children want to be taken seriously and have their viewpoints considered (Britto et al., 2004). Tools
for professionals that contribute to the overall quality of care for the child with a long-term condition are important, which could be used as a valuable addition in the care (Bergman et al., 2015; Sattoe, Hilberink, Peeters, & van Staa, 2014). Sometimes, children may want to share their results only with the healthcare professional, without parents listening. This finding is not surprising, since older children need to have confidentiality during the encounter. It has been found that adolescents describe that the encounter is impacted by the absence or presence of their parents, and that the number and subject of health topics discussed matter, since more topics are discussed when encounters are partly confidential (Gilbert, Rickert, & Aalsma, 2014). Healthcare professionals describe that the co-operation with parents are largely influencing their decisions whether to include children or not in the consultation. Still, the support is strong to include children and their preferences for participation (Coyne, 2008; Vaknin & Zisk-Rony, 2011). Indeed, it is important to also include the family in the care, as the family is essential in the child’s life (Ranerup et al., 2014). Nevertheless, if a therapeutic alliance shall occur, healthcare professionals need to take into account the child’s role as well as the parents’ role during consultations (Callery & Milnes, 2012). Therefore, children should be given a voice and choice regarding decisions about their care and whether parents should be present or not.

**Child involvement**

Depending on the approach healthcare professionals used, children got involved to a greater or lesser degree during the consultation, which was shown in the results of this thesis. An instructing approach is when the healthcare professional has their focus on the assessment tool, without exploring the items in each dimension of the web-DISABKIDS. This means that professionals gave the child a summation of the results without exploring the details behind the child’s answers. Others have also found that professionals often dominate the dialogue with the child, and this points to the need of being attentive to the child’s own narrative and what the child wants to talk about (Golsäter, Lingfors, Sidenvall, & Enskär, 2012). If healthcare professionals wish to understand and respond adequately to the child and give them a sense of being understood,
healthcare professionals need to attend simultaneously to all dimensions of illness (biological, psychological and social). It is important to encourage children to communicate in different ways, which also can help professionals to identify subgroups with impaired health, and can provide professionals with appropriate knowledge to support children concerning both treatment and self-management (Lindfred, Saalman, Nilsson, Sparud-Lundin, & Lepp, 2012). Child involvement can be understood as transitory and dependent on several factors. Children need to receive appropriate knowledge for increasing their understanding and ability to be involved. Healthcare professionals need to work together with the child, by promoting involvement through open discussions and negotiations. Moreover, healthcare professionals have to reflect on their control and power which have an effect on the caregiving (A. Davies & Randall, 2015). It could be argued that a system-oriented healthcare professional is conscious of the responsibilities to the child and the family or significant others. Therefore, the biopsychosocial model may provide healthcare professionals with the capacity to be more skilful when it comes to psychosocial areas (Engel, 1981). In addition, the delivery of healthcare for managing long-term conditions needs attention to the importance of active patients working together with a prepared healthcare professional (Wagner et al., 2001). This is a core aspect in the co-production of healthcare delivery and postulates the importance of interactions within the healthcare system as a whole (Batalden et al., 2015) as well as between children, parents and the healthcare professionals. For a high quality consultation to occur, healthcare professionals need to change their operating procedures according to the child’s needs during treatment since a flexibility in the care with regard to the timing is necessary (Wiman, Wikblad, & Idvall, 2007).

Interactions between the child and the healthcare professional are more likely to be productive if both parties are prepared by having the relevant knowledge, skills and confidence to make the best use of the encounter (Wagner et al., 2001). Healthcare professionals should also have sufficient time and resources to act, to ensure effective management in clinical practice (Wagner et al., 2001). This could be understood as a need for professionals to change stance, from objective detachment to a more reflective participation and co-productive manner in the encounter. The
application of the biopsychosocial model can be seen in the light of the need of each child. By moving beyond the multidimensional thinking, it could be recognized that participation in care is rather complex, which highlights that it is impossible to know about all contributors and influences on a specific outcome (Borrell-Carrió et al., 2004). Building a system of interaction between the healthcare professional and each child could potentially shift the use of knowledge through an active engagement from the child as a partner who gives and receives as much as the healthcare professional. This demands a movement away from profession-centredness, towards viewing care as a transformation of input to a service and to view care as linking knowledge and needs (Batalden et al., 2015). This requires an attitude change among professionals and children as well as educators and policy-makers (von Thiele Schwarz, 2016). The relationship between the child and professional should be built on trust and understanding for each other’s different expertise and knowledge. When the child has answered the questions prior to the consultation, they contribute by sharing information about their health. Consequently, the healthcare professional ought to use this information in the dialogue with the child—built on the interaction with the child—by inviting and engaging the child in the conversation.

The results point to the conclusion that a co-productive care may be facilitated when children have the opportunity to be involved and engaged in conversations about their HRQOL results. The case in the instructing approach was a simple use of the assessment tool when comparing to descriptions made by the children. This means that the use of decision supports based on HRQOL instruments will not automatically lead to better care for the child. If the outcome in using HRQOL instruments are to achieve an active child involvement and an open communication during the encounter, the use of such instruments needs to be practised and discussed in a wider perspective. For instance, focusing the care on the child’s physical and emotional needs, as well as educating professionals with the appropriate knowledge in using instruments as a base for health-related discussions (Kitson, Marshall, Bassett, & Zeitz, 2013).
Synthesis of the results in relation to the healthcare system

The results indicate that children and healthcare professionals can engage as co-productive partners in healthcare and create a structure for shared activity which go beyond the boundaries for the clinic (Batalden et al., 2015). Further, the redesign of specific elements of long-term care delivery can lead towards improvements in the quality and outcomes of care (Crandall et al., 2012), when an instrument of HRQOL is used. Healthcare should be viewed as shared work, by seeking and using all stakeholders’ expertise and by creating a sense of solidarity by promoting the idea that every person can make a difference. Aligning the child, the healthcare professional and the researchers together to motivate and increase expectations for performance could deliberately stimulate solutions (Margolis, Peterson, & Seid, 2013). A co-productive partnership is hindered or facilitated by many forces from different levels in the healthcare system and in the wider community. Understanding the aim of healthcare services and the responsibilities from each person and their role is important. A challenge is that the partnership between children and their families and the healthcare professional is dynamic and the degree of agency shifts across settings, time and circumstances. The system of healthcare cannot abandon those who do not have the resources to partner effectively in the co-production of care. Co-productive care also challenges standardisation, and the healthcare culture could be an obstacle if it is resistant to new approaches (Batalden et al., 2015). It is necessary to consider new ways in the redesign of healthcare—by preparing healthcare professionals and children and their families—in finding new organizational forms and structures for the delivery of healthcare, as well as utilising new technologies in the healthcare infrastructure.

Healthcare professionals need the opportunity to develop their competencies in using new technologies as an integrated part in the encounter. Technologies such as decision supports could be effective when they are used as a supplement during the patient encounter (Coulter & Ellins, 2007). Still, the use of information and technology systems for support in healthcare management does not bring a person-centred approach by itself. Therefore, the purpose of the support needs to be
clarified, and different forms of dialogues are required (Sparud-Lundin et al., 2013). To achieve a reconceptualization of health which is associated with a biopsychosocial view, healthcare professionals need to be open to new ideas and the implications of new technology as well as use of the supportive systems such as the quality registry, which is offered in a modern healthcare system. In addition, the ability to analyse the implications in clinical practice and the opportunity for the child with a long-term condition to share their thoughts and feelings about their health during consultations (Hwu et al., 2001) is necessary. The biopsychosocial perspective grounded in the ICF-CY may help the understanding of human functioning in everyday life as well as about the health of children with long-term conditions by addressing mental, physical, psychosocial and environmental aspects. It is further emphasized that active engagement between the child and the healthcare professional can lead to outcomes such as stable perceptions of subjective well-being and meaningfulness (Imms et al., 2016). The combination of resources from professionals and children is needed to achieve the best possible outcome which will vary depending on the condition and the person itself. Concurrently, it is necessary to use appropriate tools and measures delivered by information technology to enable the creating, sharing, shaping and applying of knowledge where healthcare takes its place (von Thiele Schwarz, 2016).
Conclusions

The results of this thesis points to the conclusion that:

DISABKIDS is appropriate to use when measuring HRQOL in children with long-term conditions, but both generic and disease-specific items are needed to illustrate a biopsychosocial perspective on children’s health.

National quality registries can play an important role if patient reported outcomes are included, since HRQOL in subgroups differentiate across several dimensions. This may be helpful when target interventions are planned. HRQOL instruments can also be used as a complement to biomedical variables (such as HbA1c) to receive a biopsychosocial perspective on children’s health.

According to children’s statements in this thesis, they request feedback on their results after answering to DISABKIDS. This implies that healthcare professionals need to prioritize time for discussions about results from HRQOL instruments during the patient encounter. Misinterpreted answers could be corrected during conversations, if healthcare professionals involve the child and use an inviting or engaging approach.

It is a benefit for children when they have the opportunity to share their results with their healthcare professional, since children may understand more about their own health. If the perspective is that everyone has potential for good health, healthcare professionals need to develop skills to mobilise environmental and personal resources among children with long-term conditions. A tool for recognizing this perspective could be to use an HRQOL instrument as a starting point for further conversation during consultations.

Sharing the results from HRQOL instruments are important, but needs to be addressed carefully during encounters. This means that the conversations between the child and the healthcare professional must be based on open-ended questions, and cannot be excluded when a HRQOL instrument is used in clinical practice. A co-production of healthcare could
be facilitated when children get the opportunity to be involved and engage in conversations with the healthcare professional.

Implications for practice

HRQOL instruments should be used in clinical practice to promote children’s knowledge concerning their health as well as promote the child’s involvement during the conversations with the healthcare professionals. HRQOL instruments can also be a complement to biomedical variables to generate a biopsychosocial perspective on the child’s health.

When information technology is used as a facilitator in the co-production of healthcare, pre-visit planning could be applied, if children get an opportunity to answer to the questionnaire prior to the visit. Robust information technology support should be incorporated in the healthcare system organization and structure. This could facilitate data collection online, where children also can answer from their home.

HRQOL instruments should be easy to access and providing sample information on the interpretation and use. It should be a multidisciplinary approach towards the use of HRQOL instruments in clinical practice. For example, integrating HRQOL results in existing team meetings, which may facilitate the possibility to design and plan care.

A system for collecting HRQOL data should be flexible according to the clinical situation. This means, that the organisation must bear the responsibility for coordination and continuing the process of implementation. Healthcare professionals need time to learn and practice new competencies regarding the use of technology such as quality registry systems and decision supports. This implies that an extensive development in the healthcare infrastructure is needed to design and specify the appropriate way to use this technology.
Implications for further research

The bridge between research findings and the implementation in clinical practice is still rather scarce. Therefore, methods to study and evaluate how to implement research findings are needed, for example by using collaborative learning networks. The use of clinical interventions could be further developed and studied in more detail, which may be helpful in the implementation of research findings.

In this thesis, the instrument of HRQOL was based on a former questionnaire, but with modern technology better questionnaires with adaptive testing could be developed and their applicability in a clinical context should be studied.

What matters to children differs across gender, age, as well as cultural background, therefore, focusing on the healthcare service and what is important for different groups, including children with different ethnical backgrounds needs to be studied.

Although there is evidence about using patient reported outcomes and giving feedback to children, there has not been any attempt to synthesize this evidence. Qualitative studies can help clarify the barriers and facilitators to change and the obstacles in complex interventions. Such studies could provide a deeper understanding into the use of patient reported outcomes in the future.
Svensk sammanfattning

Introduktion

I Sverige finns närmare 2 miljoner barn under 18 år och cirka 20 % av dessa barn har en långvarig sjukdom. Med en långvarig sjukdom menas att barnet behöver vård, uppföljning eller medicinering i mer än tre månader samt att tillståndet inte kan läka spontant eller går att bota. När barn beskriver hur de upplever att leva med en långvarig sjukdom framkommer bland annat att de kan ha svårigheter att delta i fritidsaktiviteter, eller att de har en känsla av att vara annorlunda, vilket kan leda till nedstämdhet och oro. Det har beskrivits i studier att barn kan ha svårt att uttrycka sina tankar och känslor till vårdpersonal vid besök i hälso- och sjukvården. Studier har också visat att det ofta är vårdpersonal som för samtalet vilket kan medföra att barn inte kommer till tals kring frågor som är viktiga för dem. Ett sätt att underlätta för barnen är att de får möjlighet att delge information om hur de mår genom att använda så kallade patientrapporterade mått. Det innebär att barnen svarar på frågor om hälsorelaterad livskvalitet med utgångspunkt i strukturerade frågeformulär före sitt besök i hälso- och sjukvården. Detta har testats internationellt i studier med barn med diabetes, reumatism och cancer som visat att psykosociala frågeställningar blir tydligare under samtalet mellan barn och vårdpersonal när sådana frågeformulär används.

Sedan mitten av 1970 talet har det i Sverige utvecklats nationella kvalitetsregister. Registren har som främsta uppgift att säkerställa god kvalitet och följa upp vård och behandling för olika sjukdomsspecifika grupper. En vision för kvalitetsregistren är att de ska användas integrerat och aktivt för lärande, förbättring och forskning för att tillsammans med personen skapa bästa möjliga hälso- och sjukvård. Ett sätt att möjliggöra denna vision är att inkludera patientrapporterade mått, där personen själv kan dela sin erfarenhet av att leva med en specifik sjukdom. Därför föll det sig naturligt att i denna avhandling använda strukturen för nationella
kvalitetsregister för införandet av patientrapporterade mått för barn och ungdomar med långvariga sjukdomar.

Det övergripande syftet med denna avhandling var att utforska och beskriva hur frågeformulär för hälsorelaterad livskvalitet kan användas i uppföljning av barn med långvarigt hälsotillstånd genom att utgå från systemet med nationella kvalitetsregister. I den första delstudien var syftet att beskriva innehållet i olika typer av frågeformulär utifrån ett biopsykosocialt perspektiv. Den andra delstudien syftade till att beskriva skillnader i hälsorelaterad livskvalitet hos barn med typ 1 diabetes. I delstudie tre var syftet att utforska och beskriva barnens erfarenheter av att besvara frågeformulär i anslutning till sitt mottagningsbesök. I den avslutande delstudien studerades hur vårdpersonal använder sig av dessa frågeformulär i mötet med barnen.

Metoder


I denna avhandling har totalt sex olika frågeformulär granskats. Det var 204 barn som tackade ja att besvara DISABKIDS och 131 frågeformulär returnerades (svarsfrekvens 64 %). Därefter intervjuades 25 barn i anslutning till deras mottagningsbesök och intervjuerna ljud-upptogs och transkriberades sedan ordagrant för att sedan analyseras med induktiv innehållsanalys. Totalt 21 mottagningsbesök videoinspelades där sammanlagt nio olika läkare och sjuksköterskor använde sig av
DISABKIDS resultatet i samtal med barnen. Filmerna analyserades också med induktiv innehållsanalys.

Resultat

mötter det barnen efterfrågar. Det betyder att användningen av ett frågeformulär för hälsorelaterad livskvalitet inte automatiskt genererar en bättre vård för det enskilda barnet, utan beror på hur instrumentet används under samtalet.

**Slutsatser och rekommendationer**

Acknowledgements

I would like to thank Futurum – the Academy for Health and Care, Region Jönköping County, Jönköping, Sweden and the Ebba Danelius foundation at Swedish Society of Nursing for financial support, which made this research possible. I also want to express my appreciation to all children and healthcare professionals who voluntarily participated in the studies of this thesis, without you, there would not be any thesis.

There are many people who have supported me which have made this research possible and to whom I would like to express my sincere gratitude. First, I would like to thank my four supervisors: Professor Karin Enskär, my main supervisor, thank you for believing in me, since I often have had my own doubts. Your knowledge and ideas have no limits, and we have had a lot of interesting discussions. Professor Boel Andersson Gäre, I admire you and your energy and your deep knowledge in quality improvement. Thank you for sharing that. Karin Åkesson, MD, associate professor at Linköping University, thank you for your valuable contributions about the quality registry and your deep knowledge about children and diabetes. Karina Huus, associate professor at School of Health and Welfare, thank you for helping me in the “fishing expedition”. We have had many laughs. I would also like to thank other co-writers of my papers in this thesis. Marie Golsäter, PhD your knowledge in analyzing video-recorded data is amazing. Thank you for the walks and talks with views over the beautiful landscapes of the Spanish islands. Professor Rune J Simeonsson, University of North Carolina, Chapel Hill, USA, thank you for all your readings and new ideas when we wrote paper I. I would also like to thank you for all help we had during my stay at UNC and Chapel Hill, for dinners, conversations and that extra bed for our son. I would also like to thank associate professor Ulf Samuelsson, MD and PhD and Lena Hanberger, PhD at the University of Linköping for support during the data-collection for paper II.

Another special thanks to professor Mats Granlund and professor Ewa Björck Åkesson, CHILD, because you gave me the opportunity to be a scholar at the program of Transatlantic Consortium on Global Education
and Developmental Studies (GEDS) which took me and my family to UNC and Chapel Hill, USA.

I would also like to express my sincere thanks to Per Lewander, MD and Bo Magnusson MD, for sharing your clinical expertise about kidney diseases and rheumatology, and new ideas of thinking about quality registry. I admire your work and never ending engagement for making the care for these children better. Also, thanks to associate professor John Chaplin, University of Gothenburg, for sharing knowledge about the DISABKIDS questionnaire and patiently answering my questions.

I appreciate all my fellow students, past and present at the Research School of Health and Welfare, Jönköping, for the time we have shared and discussions we have had, from scientific theories and methods to the winner of the Eurovision song contest. A special thanks to the coordinator Kajsa Linnarsson, for support and happenings we have had. A special thanks to professor Ewa Idvall, Malmö University, Dan Malm, associate professor at the School of Health and Welfare and PhD Student Annika Nordin, for reviewing my thesis during the final seminar. Also, a very special thanks to Peter Batty, for your time and proof-readings of my texts in this thesis. Gunilla Brushammar, Jönköping University Library, thank you for your expertise and support with the references and EndNote support.

I am also fortunate to have many good friends who have encouraged and supported me. Thank you for all the walks and coffee-breaks and dinners, when you have listened to me and “my crazy work”. A special thanks to Elisabeth Gustavsson, Tina Samefors, Ammi Lindrud and Emelie Terlinder. I still cannot believe that one of you actually read this thesis! Last, but not least, my husband Henrik – thank you for preparing breakfasts and dinners. You are the foundation of our family, my best friend and you know me better than anyone. Thank you for all the support and unconditional love, when listening to my doubts when it has been difficult. My beloved children, Agnes, Signe and Edvin, for all the love and encouragement you give me every day. And Signe, you and only you know how it is, thank you for putting up with all my questions all the time!

Jönköping December, 2016
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