Juvenile chronic physical illness in Northern Russia
Studies on mental health, health-related quality of life, and family functioning

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You are braver than you believe,
stronger than you seem,
and smarter than you think

A. A. Milne, Winnie the Pooh
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Paper I - IV
ABSTRACT

Background
Chronic physical illness (CPI) is a prolonged, rarely cured condition, which often causes impairment of activities of a child’s or adolescent’s daily living. This thesis encompasses three cohorts of patients with CPI – diagnosed with diabetes, asthma or epilepsy. Psychological disturbances and difficulties experienced by young patients with CPI are common, and changes in the family environment are inevitable. Hence, from a health care perspective, three cohorts of CPI can give a frame of reference to guide our understanding on the psychological health of adolescents with CPI, and the disease impact on their life; to determine target groups for psychological interventions, and to identify important directions for health care development.

Objectives
The overall aim of the study was to assess the psychological well-being of adolescents with CPI in Northern Russia, as well as to identify factors of potential risk or protective significance for adolescents’ well-being.

Methods
The questionnaires covering different aspects of psychological well-being as behavior-emotional problems, depressive symptoms, self-esteem, and health-related quality of life (HRQoL) as well as family functioning were answered by the adolescents with CPI and their mothers, including the Child Behavior Check-List (CBCL) and Youth Self-Report (YSR); Beck Depression Inventory (BDI); The Rosenberg Self-Esteem Scale; I think I am; Quality of Life in Epilepsy Inventory for Adolescents (QOLIE-AD-48); Diabetes Quality of Life Questionnaire for Youths (DQOLY); Pediatric Asthma Quality of Life Questionnaire (PAQLQ); Self-report Family Inventory (SFI); and Socio-Economic Status (SES) questions. Disease related clinical information was withdrawn from the pediatric outpatient clinic. The sample consisted of 148 adolescents with CPI identified from Arkhangelsk pediatric outpatient clinic records and their mothers. Comparative data were obtained from a group of 301 schoolchildren and their mothers.
Results

Results in Paper I showed certain differences in mental health of adolescents with CPI compared to healthy counterparts indorsed by mother’s reports alone, while self-reports by adolescents with CPI didn’t show any discrepancy in the level of behavior/emotional problems and depression compared to healthy peers. Most pronounced symptoms were found in adolescents with asthma and epilepsy associated with disease severity and gender. In the results of Paper II there were no significant differences found in self/mother reported family functioning of the total group with CPI versus controls. The perception of family functioning differed between the CPI groups and was associated to certain disease-related and non-disease factors. Paper III showed that adolescents with diabetes and asthma maintain positive self-esteem similar to or even higher than that of their healthy counterparts. Adolescents with epilepsy reported lower self-esteem compared to other CPI groups or controls. A diversity of factors contributed to self-esteem variation within the CPI groups. Paper IV provides evidence that adolescents with CPI maintained relatively moderate to high levels of HRQoL. The domains affecting HRQoL were related to both disease-specific (severity) and non-disease factors (gender and SES).

Conclusions

In the scope of the present thesis we assessed and described psychological well-being and family functioning of adolescents with chronic illnesses. Our results demonstrate generally low levels of behavior-emotional problems, depressive symptoms, and relatively high self-esteem, positive health-related quality of life, and successful family functioning in adolescent with CPI in Northern Russia. However, we conclude that there is a risk of mental health problems, particularly of internalizing nature, in adolescents with asthma and epilepsy. The significant risk factors associated with psychological well-being mediated by family functioning in CPI groups were single-parent household, child age, disease severity, child gender, family SES, and changes in the family life. High family competence and successful family functioning are indisputable protective factors for psychological well-being in adolescents with CPI.

Key words: chronic physical illness, adolescence, mental health, behavior/emotional problems, depression, self-esteem, health-related quality of life, family functioning
LIST OF PUBLICATIONS

This thesis is based on the following original papers, which will be referred to by their Roman numerals.


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ABBREVIATIONS

AED  Anti-Epileptic Drugs
AMOS Analysis of Moment Structures
ASEBA Achenbach System of Empirically Based Assessment
BDI Beck Depression Inventory
CBCL Child Behavior Check-List
CNS Central Nervous System
CPI Chronic Physical Illness
DQOLY Diabetes Quality of Life Questionnaire
GLM General Linear Model
HbA$_{1c}$ Glycosylated hemoglobin
PAQLQ Pediatric Asthma Quality of Life Questionnaire
QOLIE-AD-48 Quality of Life in Epilepsy Inventory for Adolescents
RSES Rosenberg Self-Esteem Scale
SEM Structural Equation Modeling
SES Socio-Economic Status
SFI Self-Report Family Inventory
YSR Youth Self-Report
WHO World Health Organization
1. INTRODUCTION

As many as one child in six is affected by chronic physical illnesses (CPI) (van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). By CPI we understand a disease of long duration and generally slow progression. Pediatric care dealing with chronic conditions has been developing extensively during last 3-4 decades. Most of these children now survive for longer time, the longer life expectancy is often associated with lifelong health problems. Therefore, more may suffer psychological disturbances and difficulties (Perrin, Gnanasekaran, & Delahaye, 2012). Three chronic disease groups with significant prevalence across the world and in Russia were chosen for this study: type 1 diabetes, asthma, and epilepsy. According to the conclusion of Sabgaida and Okunев in a recently published study, the incidence rates of these three diseases in the child and adolescent population in Russian Federation are constantly growing from 1992 to 2010 (Sabgayda, 2012). The latter epidemiological study used the only available source of information on prevalence and incidence rates of morbidity in the population, a certain statistical form filled by each medical care unit in the country. The incidence of endocrine diseases in adolescents increased 3.1 times, while the incidence in younger children 2.1 times. The incidence of the respiratory diseases in children has also grown substantially, as well as the incidence rates of diseases involving nervous system (Sabgayda, 2012).

The three chronic diseases included in our study have certain biological similarities and differences each posing distinct psychosocial challenges. According to Rolland and Walsh’s classification these three CPI are nonincapacitating (Rolland & Walsh, 2006). Type 1 diabetes on average has a gradual-onset (although life threatening if not treated) and life-long duration, while asthma and epilepsy have a more acute and relapsing form. Although different, these three diseases share the following features: unpredictable course, necessary behavioral adjustments, long-term dependency on treatment, and strict commitment to self-management strategies (Kyngas, 2000). Crucial for psychological impact is also outcome expectancy and level of uncertainty related to the illness.

In spite of the fact that young people with CPI account for a large portion of the work of pediatricians and other medical professionals, the need to identify psychosocial issues is often unrecognized by medical specialists. The primary health care physicians receive limited training in the psychosocial issues related to chronic disease, despite the acknowledgement that these factors are important determinants of course and outcome (Geist, Grdisa, & Otley, 2003).
CPI management in children is a problem that besides pediatric health care quality combines elements of economy, demographics, culture, education, and therefore it concerns the whole society. The health status of children is an indicator of the status and well-being of society as a whole. WHO describe “health” - as a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity.

One of the most important priorities of the present is to create the necessary conditions and sufficient measures for the adequate CPI perception, smooth adjustment to CPI, and successful management of the disease by adolescents. Developing the healthcare system with the emphasis on medical, psychological and educational rehabilitation may help to improve children’s health and their social adaptation.

As mentioned above, psychological well-being in children and adolescents with chronic disorders was studied repeatedly during the last decades. The project behind the present thesis was initiated due to the lack of information on the subject from the studied area. The research conducted and published in Russia and in Russian language is available for a small number of readers from international research community, although the interest in the topic has been growing. According to the World Bank standards Russia is viewed as a developing country. Because the majority of the world’s children and adolescents live in the developing rather than developed countries, engaging youths growing up in a developing world in research is important (Grigorenko, Geiser, Slobodskaya, & Francis, 2010).

1.1 Type 1 diabetes

Type 1 diabetes (previously known as insulin-dependent or childhood-onset diabetes) is a metabolic disorder of complex etiology involving genetic and environmental factors (Eringmark Regnell & Lernmark, 2013), and characterized by chronic hyperglycaemia with disturbances of carbohydrate, fat and protein metabolism which are connected to lack of insulin production (Malik & Taplin, 2014). The outcome of type 1 diabetes includes long–term damage, dysfunction and failure of various organs (WHO 1999).

The global variation in the incidence of type 1 diabetes in children and adolescents is well-known. The overall age-adjusted incidence of type 1 diabetes varied from 0.1/100,000 per year in China and Venezuela to 36.8/100,000 per year in Sardinia and 36.5/100,000 per year in Finland (Karvonen et al., 2000). The incidence in Russian Federation in 2009 as reported by Shiryaeva was 15.66 per 100,000 of child population (Shiryaeva, 2009). In Arkhangelsk region the incidence rates are similar to the North-West Federal District figures – 15.7/100,000 for 2008 (FRIHOI 2014).
The prevalence of type 1 diabetes in Russian Federation increased from the year 2000 to 2009 in children by 35.7% (from 59.4 to 80.6 per 100 thousand child population), and in adolescents by 68.9% (from 108.5 to 183.5 for 100 thousand teenage population) (Suntsov Yu, 2011).

Starting in 1995 the federal target program “Diabetes” was adopted in Russian Federation. It aimed to increase the duration and quality of life by providing specialized medical care for patients with diabetes, preventing diabetes, and reducing its late complications involving sufficient financial resources.

Diabetes has for a long time been one of the main priorities for the health care system in Arkhangelsk region. The new concept of juvenile diabetes care has been applied from 1995 when nationwide program “Diabetes” was introduced. Many aspects of life with diabetes were addressed: easy access to care, free medication, means of insulin administration and blood sugar control, “the family as a patient” approach, involvement of the patients and their parents in activities aiming to increase the quality of life of children with diabetes. This approach led to the improvement of pediatric care and treatment of diabetes compared to somewhat extensive and rigid Soviet approach to chronic disease management.

Glycosylated hemoglobin, another name for HbA1c, is an objective value of average blood glucose concentration over approximately two previous months and should be measured regularly, usually every 3 months. Although the ideal goal is to achieve an HbA1c value as close to normal as possible, goals that most patients should achieve vary with age: 7.5% to 8.5% for toddlers and preschoolers (<6 years), less than 8% for school-age children (6 to 12 years), and less than 7.5% for adolescents and young adults (13 to 19 years) (Cooke & Plotnick, 2008). However, hypoglycemia often limits the ability to achieve these goals. Indeed, with the limitations of current insulins and methods of administration, such target levels often cannot be reached, especially in adolescents. In Russian Federation about 92.6% of people with type 1 diabetes have the level of HbA1c >7.5%, and only 12.6% have HbA1c <7.0% (Suntsov, 2011).

1.2 Asthma

Asthma is a chronic disease characterized by recurrent attacks of breathlessness and wheezing, which vary in severity and frequency from person to person. During an asthma attack, the lining of the bronchial tubes swells, causing the airways to narrow and reduce the air flow, leading to expiration problems. In children asthma is often of allergic nature with a complex etiology with gene-environment interaction (Subbarao, Mandhane,
Asthma is the most common chronic disease among children (WHO 2013).

In the Arkhangelsk region, the prevalence of asthma is approximately 11 cases per 1000, which is close to the world’s average numbers. Statistics of the prevalence of asthma is usually based on figures obtained from the patient’s referral rates to medical facilities, not taking into account children who for various reasons are not referred to the hospital or outpatient clinic. Thus, epidemiological studies carried out using different methodological approaches, suggest that the true prevalence of asthma among children in Russia is several times higher than the official figures (Lyapunova, 2009). For instance the study carried out in two central districts of St. Petersburg reported 7.4% - the prevalence of childhood asthma which is many times higher than what is registered in the medical records (0.8%) or presented in the official data (1.5%) (Glushkova & Grjibovski, 2008). Asthma prevalence has been shown to increase also in other countries, e.g. a recent population-based Norwegian study in a northern county of Norway showed increasing prevalence of asthma ever (7.3% in 1985 to 17.6% in 2008) among children of 7-14 years of age (Hansen, Evjenth, & Holt, 2013).

In 2008 the registered incidence rates of asthma in child population of Arkhangelsk region was 130.4 per 100,000, which is lower than the average numbers for Russian Federation 170.5 per 100,000 (FRIHOI 2014). Although the prevalence of asthma in Arkhangelsk region is not extremely high, it is constantly growing. Due to this fact, during the last decades disease-specific asthma care units were established. In 1998 the Asthma Center was created in Arkhangelsk regional hospital and by 2002 in Arkhangelsk there were 8 asthma schools (for adults and children). The purpose of training at asthma schools is to provide the patient/patient’s caregiver with necessary information, practical skills, methods of self-control and ability to adjust their medication in accordance with the plan developed by the physician.

1.3 Epilepsy

Epilepsy is a disorder characterized by recurrent seizures, which may vary from a brief lapse of attention or muscle jerks, to severe and prolonged convulsions. The seizures are caused by sudden, usually brief, excessive electrical discharges in a group of neurons. The typology of the seizure depends from the area involved at the onset and on the area mobilized in the spread of the discharge. Such symptoms of neurological dysfunction do not necessarily represent a condition known as epilepsy, which is the term used for recurrent seizures which arise apparently spontaneously, thereby excluding such conditions as hypoglycemia or hypocalcaemia (Dreifuss,
The seizure classification is based on the objective documentation of individual seizure types divided into focal seizures, generalized seizures, and unclassified seizures (Berg et al., 2010). The epilepsies are recognized as electroclinical syndromes – a cluster of clinical symptoms and electroencephalographic characteristics that appear at certain ages (Chen, 2011). Comorbidity is common in childhood epilepsy as developmental delay, autism, ADHD, depression and other chronic medical conditions (Baca, Vickrey, Caplan, Vassar, & Berg, 2011).

In a recent population-based epidemiological study conducted in Italy the reported prevalence of active epilepsy was 6.62 per 1000 (Cossu et al., 2012). The incidence rates of epilepsy for 2008 in Russian Federation was 71.3/100,000, and in Arkhangelsk region 66.3/100,000 of child population (FRIHOI 2014). A population based study in children from a Swedish county reported the prevalence of 3.5/1000 (Larsson & Eeg-Olofsson, 2006). The 10-year outcome was fairly good as 75.6% of patients were in remission (Jonsson & Eeg-Olofsson, 2011).

Recent studies in developed and developing countries have shown that up to 70% of newly diagnosed children and adults with epilepsy can be successfully treated (i.e. their seizures completely controlled) with antiepileptic drugs (AEDs). After two to five years of successful treatment, drugs can be withdrawn in about 70% of children and 60% of adults without relapses (WHO 2013).

Epilepsy is one of the world's oldest recognized conditions. Fear, misunderstanding, discrimination and social stigma have surrounded epilepsy for centuries. Some of the stigma continues today in many countries and can impact the quality of life for people with the disorder and their families (WHO 2013).

Spontaneity and unpredictability of seizures, lack of self-control during the attack causes the negative societal attitudes towards patients with epilepsy leading to stigmatization of patients. People with epilepsy experience reduced access to health and life insurance, a withholding of the opportunity to obtain a driving license, and barriers to enter particular occupations, among other limitations. In many countries legislation reflects centuries of misunderstanding about epilepsy. In both China and India, epilepsy is commonly viewed as a reason for prohibiting or annulling marriages. In the United Kingdom, a law forbidding people with epilepsy to marry was repealed only in 1970. In the United States, until the 1970s, it was legal to deny people with seizures access to restaurants, theatres, recreational centers and other public buildings (WHO 2013).

Rights of patients with epilepsy in Russia are determined by existing legislation, as well as numerous departmental instructions often based on
outdated standards. Clearly defined are the limitations on employment for military service, fire protection, public transport, navy, driving. In Russia a person with diagnosed epilepsy can't be admitted to medical school (or work with patients as a medical doctor or nurse), pedagogical school (or work as a teacher).

It should be noted that disease specific characteristics as epilepsy subgroups or comorbidity in epilepsy are not in the focus of the present thesis.

1.4 Psychological well-being in CPI

In the studies of the present thesis, psychological well-being will be regarded as a construct incorporating individuals'/parents' perception of mental health with respect to behavior-emotional problems, depression, self-esteem, and HRQoL.

CPI can have significant psychological and social consequence for the adolescent and the adolescent’s family. Many studies report maladjustment, the increase of behavior-emotional problems, depression, and decreased levels of self-esteem in this population.

The turmoil of adolescence aggravated by CPI can cause a negative attitude not only to the disease itself, compliance with the treatment and changes in the routines, but to the perception of oneself, the family and social environment. In adolescence CPI can be challenging regarding the responsibility for one’s drug/insulin intake, other treatment procedures, exercise needs and limits, and the disease management in general. The social acceptance of the disease becomes more important in adolescence, as well as the appearance, physical activity and achievements. Transition through adolescence can compromise school performance in youths with CPI due to hospitalization and time spent on disease management.

The Pinquart and Shen meta-analysis published in 2011 reported elevated levels of behavioral problems in children and adolescents with CPI, with the stronger elevations of internalizing problems, social problems, attention problems, and thought problems than of externalizing problems (Pinquart & Shen, 2011b). The type of the CPI disease in adolescents have been found to be the strongest predictor for depression; when controlling for age, gender, and disease duration (Rao, Ramu, & Maiya, 2011). Pinquart meta-analysis based on more than 600 studies found that children with CPI have lower levels of self-esteem than their healthy peers or test norms, although most differences were small. The strongest reductions were observed in adolescents rather than children, females, children from developing or threshold countries rather than developed countries, observer rating rather than child reports (Pinquart, 2013).
Psychological and social consequences of CPI in children and adolescents are also revealed in self-reported HRQoL - a subjective aspect capturing the general effect of the illness on everyday living. The importance of assessing HRQoL was emphasized both in research trials and clinical practice to evaluate the impact of the disease on child's life and to ensure that children benefit from treatment (Golden, 1998; Juniper, 1997). Deleterious effect of CPI on HRQoL has been a discussion topic in many studies (Montanaro, Battistella, Boniver, & Galeone, 2004; Sawyer, Spurrier, Kennedy, & Martin, 2001).

1.5 Analysis parameters

The present thesis covers two main research topics – psychological well-being and family functioning in adolescents with CPI.

1.5.1 Psychological well-being

A. Mental health

WHO defines mental health as a state of well-being in which every individual realizes his or her own potential, can cope with the normal stresses of life, can work productively and fruitfully, and is able to make a contribution to her or his community (WHO 2013). Mental health is an integral and essential component of health. The positive dimension of mental health is stressed in WHO's definition of health. Good mental health allows for cognitive and emotional flexibility, which are the basis for social skills and resilience in the face of stress. This mental capital is vitally important for the healthy functioning of families, communities and society (Varavikova, 2002). Child and adolescent mental health and well-being principals are well accepted by practice and research worldwide. The substantial body of research is focused on behavior, emotional, and social well-being of children and adolescents with CPI as key components of mental health. Different prevalence figures of mental health problems were reported depending on the measure, method and CPI characteristics.

This thesis makes an emphasis on three aspects of this gross parameter - Behavior-emotional problems, Depression, and Self-esteem.

a. “Behavior-emotional problems” is a parameter reflecting the assessment of diverse aspects (social, behavioral and emotional) of adaptive or maladaptive functioning of children and adolescents. Achenbach identified by means of second-order factor analysis of syndrome scale two main categories of behavior-emotional problems in children – externalizing and internalizing problems. The Internalizing grouping mainly reflects problems within the self, such
as anxiety, depression, somatic complaints without known medical cause, and withdrawal from social contacts. The Externalizing grouping, by contrast, represents conflicts with other people, aggression, and rule breaking behavior. The highest level in the hierarchy of the scores obtained from ASEBA forms comprises the Total problem scale (Achenbach & Rescorla, 2001). The syndromes completing each problem category are shown in Figure 1.

![Figure 1. Youth-Self Report, broadband and narrowband subscales (T.M Achenbach, 1991b).](image)

b. “Depression” denotes an illness characterized by a change in mood that is persistent and sufficiently severe for it to be labelled a disorder. There are two psychological perspectives on environmental causation of depression. First, Aaron Beck proposed that early negative experience results in an enduring triad of negative cognitions about the self, the world and the future. Being embedded as a latent negative schema, it can be activated by subsequent event. Second, the theory by Seligman and Abramson has proposed that frequent exposure to uncontrollable and unpredictable events leads to an enduring loss of adaptive behaviors (Goodyer, 2001). Many of the environmental risk factors for depression in adults are also potent in children. They cluster in two main areas: family history, and living in chronic difficult life circumstances or undergoing negative life events. Considering depression risk factors in youths,
negative cognition, low self-esteem and ineffective coping are particularly important (Beardslee & Gladstone, 2004). Gender differences in frequencies of depressive symptoms are substantial, with higher prevalence of depression among girls (G. Olsson & von Knorring, 1997). Specific risk factors were also identified for girls and boys. For boys, neonatal problems and poor health development; for girls, depression risk factors included family composition, death of the parent, and difficulties in school (Beardslee & Gladstone, 2004). Depression as a major effect of CPI was evidenced by comprehensive research (Bernstein, Stockwell, Gallagher, Rosenthal, & Soren, 2013; Rao et al., 2011).

Since the categorical diagnostic methodology of depression is beyond the scope of the present thesis, youth self-reports of depressive symptoms (BDI) were analyzed.

c. Self-esteem and self-concept are fundamental parts of child psychological functioning. These are two closely related personality constructs in psychology developing through life, reflecting the emotional and cognitive dimensions of self. Because diagnostic assessments (e.g., self-report) of these personality dimensions always cover both aspects, it may be justified to perceive self-esteem and self-concept as just one construct, reflecting the way an individual perceives and evaluates his own acceptance and/or competence (Vermeiren, Bogaerts, Ruchkin, Deboutte, & Schwab-Stone, 2004). The theoretical approach to self-esteem and self-concept is presented in Figure 2.

![Figure 2. Theoretical approach to self-assessment.](image)

The complexity of the variables related to the perception of self-esteem was presented by DuBois and Hirsch in a general conceptual model of self-
esteem in early adolescence (DuBois & Hirsch, 2000). A multidimensional structure of self-esteem in this model is influenced by a complex of individual (physical, cognitive, behavioral developmental status) and contextual (such as peers, family, and school) factors, with bidirectional linkage to adjustment outcome (Figure 3). A recently published review has shown that self-esteem in children and adolescents can be contradicted by chronic illness (Pinquart, 2013). The well-known self-esteem development patterns were observed in this study – lower self-esteem in girls than in boys, in adolescents than in children, in children from developing or threshold countries.

![General conceptual model of self-esteem in early adolescence. Debois & Hirsch, 2000.](image)

**B. HRQoL**

HRQoL is generally considered as a multi-factorial construct focusing on individuals' subjective evaluations of their physical health, mental health and social functioning (Sawyer et al., 2001) and the objective impact of the disorder associated with the disease or medical treatment (Gill & Feinstein, 1994). HRQoL is an important outcome in studies of pediatric patients with chronic physical illnesses (Golden, 1998; Juniper, 1997; Montanaro et al., 2004). Research has shown that HRQoL in children and adolescents with CPI is markedly lower than that of the general population (Montanaro et al., 2004; Sawyer et al., 2001).

There is a clear distinction between QoL, HRQoL and health status. As was suggested by Feldman et al. all three measures should be considered for use in health studies. They showed that children referred to rheumatologic care
did differentiate between overall QoL, HRQoL and health status (Feldman, Grundland, McCullough, & Wright, 2000). Results of Smith’s et al. meta-analysis in adult population showed that only two domains – mental health and physical functioning - are the key determinants of the QoL judgments, social functioning did not have a major impact on QoL construct. Their findings also emphasize that in the context of CPI, QoL is a subjective appraisal of one’s current life based primarily on psychological functioning and to the lesser degree to the physical functioning (Smith, Avis, & Assmann, 1999). In the adolescent population it could be different due to the significance of the social component in this developmental stage, and in case of our study due to the use of the disease-specific tools. The importance of the social component of HRQoL (including the relationship with peers) was emphasized in the cross-national survey exploring the HRQoL perceived by 8 to 18 year old healthy children (Detmar et al., 2006).

Disease-specific tools are designed to be applicable to specific patient populations, usually defined by disease pathology. Measurement priorities are focused on aspects of disease-specific health status that are likely to be sensitive to treatment and natural history (McHorney, 1999).

1.5.2 Family functioning

Family functioning is a broad concept and is often used as an umbrella term encompassing numerous constructs, including parents’ satisfaction with their parenting role, parent-child interactions, family communication, family adaptability, and family cohesion (Rolland, 1993). In our study we adapt Beavers family functioning concept with the emphasis on family competence. The latter relates to structure and adaptive flexibility of the system (Beavers & Hampson, 2000). Effective family functioning is a combination of both structure and the ability to change structure. According to Beavers, family health is in its flexibility, and competent families alter their style according to the developmental needs throughout the family life cycle (Beavers, 1993). Hence with the CPI diagnosis of a child, the competent family changes and adapts in various ways in order to meet child needs. On the other end of the continuum are nonfunctional families with the rigid behavior patterns and responses, with more limited freedom to evolve and differentiate. Family functioning is a key variable that has been shown to play an essential role in children’s adjustment to CPI (McClellan & Cohen, 2007).

Wallander et al. proposed a conceptual risk-resistance model predicting child’s psychological adjustment to CPI as a function of maternal and family adaptation (Wallander, Varni, Babani, Banis, & Wilcox, 1989). In the model the effects of the risk factors (e.g. specific diagnosis, severity/visibility of the condition, cognitive impairment, impaired functional independence and
psychosocial stressors) on child’s psychosocial adaptation may be attenuated by certain resistance factors (e.g. intrapersonal, social-ecological and stress-processing). The social-ecological factors include family environment, social support, parental adjustment and resources in community/culture.

1.6 Risk and protective factors

1.6.1 Socio-demographic factors

Age
The impact of CPI on children over time is a matter of discussion. Age can be a risk factor due to accumulation effect. A clear understanding of lifelong threatening disease and consequences of the complications due to maturing can be perceived negatively. As well as repeated absence from school and separation from peers due to hospitalization (Pinquart & Shen, 2011a). On the other hand, older age can have a protective significance. Reducing effects of CPI on adolescents functioning due to the maturation, adaptation to the disease itself, the treatment, the necessity of the compliance and stabilized family adjustment (Rodrigues & Patterson, 2007). The individual psychological characteristics as well the disease features can cause the diversity of the CPI perception over time. As children move into adolescence, their CPI and psychological/psychosocial comorbidities may continue or may develop for the first time. Establishment of adolescents own identity, emotional separation from parents, strong peer group identification, increased health-risk behaviors, and development of vocational plans are issues specific for adolescence (Perrin et al., 2012). Some of these issues are challenged by CPI. Taylor’s review identified seven common themes adolescents bring up when asked about living with chronic illness: developing and maintaining friendships; being normal/getting on with life; the importance of family; attitude to treatment; experiences of school; relationship with the healthcare professionals; and the future (Taylor, Gibson, & Franck, 2008).

Gender
During puberty the observed differences between genders are more obvious, regarding both biological vulnerability and social behavior (Berg-Kelly, 1999). In previous studies gender proved to have a major role in psychological adjustment, well-being and day to day living of adolescents with CPI. Research on a general adolescent population show that girls are at the increased risk for psychological disturbance (internalizing behavior problems, poorer self-concept) during the transition through adolescence
(Austin, Dunn, & Huster, 2000). According to Pinquart and Shen review boys with CPI are more likely to react toward their illness with externalizing symptoms (Pinquart & Shen, 2011b). Lower level of self-esteem in children with CPI was found in girls than in boys (Pinquart, 2013). Girls were also reported to be more vulnerable regarding the development of the emotional problems, suicide ideation, and lower HRQoL than their male counterparts (Alvim et al., 2009; Graue, Wentzel-Larsen, Hanestad, Batsvik, & Sovik, 2003; Suris, Parera, & Puig, 1996).

Socio-demographics

Social class, education/occupation, and income level of the family are important determinants of psychological well-being in children. Studies that determined the effect of SES on mental health problems reported more problems and lower competence in children with low SES background (Verhulst, 2004). There is a substantial body of research suggesting the association between psychological well-being of CPI children and socio-economic status of the family (Beardslee & Gladstone, 2004; Chen, Fisher, Bacharier, & Strunk, 2003; Erickson et al., 2002). The research by Gortmaker et al. indicated that risks of behavioral problems associated with chronic conditions are associated with low family income, low maternal education and living with fewer than two parents (Gortmaker, Walker, Weitzman, & Sobol, 1990).

A child’s chronic condition can affect family finances adversely. Families may face significant out-of-pocket costs related to the child’s condition (Perrin et al., 2012). Families from a poorer socio-economic background are particularly vulnerable, as the time demands of caring for a child with a chronic condition may force parents to stop or cut back on work hours, with implications for financial well-being (Kuhlthau & Perrin, 2001).

Family structure (number of adults and children), family type (single/two parent family), and changes in the family life (divorce/death) are other factors that can interact with the psychological well-being of children and adolescents (Beardslee & Gladstone, 2004; Perrin et al., 2012).

1.6.2 Disease related and health care associated factors

Disease severity

Disease severity is the interaction between biological severity and environmental factors including medical treatment (Stein et al., 1987). The relation between disease severity and psychological functioning is unclear. A lot of findings indicate that there is no direct association between disease severity and the level of psychological adjustment (Barlow & Ellard, 2006).
At the same time, other studies show the relationship between poor physical health and psychological well-being of young patients with CPI (Zullig, Valois, Huebner, & Drane, 2005). Disease severity was proved to exhibit negative changes in children’s psychological profile, elevate the risk of adjustment difficulties, and forecast behavioral problems (Eiser, 1990). Furthermore, the disease severity or functional severity to a certain extent was associated with a negative impact on the family functioning reported by the sample of mothers and fathers of children and preadolescents diagnosed with CPI (Rodrigues & Patterson, 2007).

**Age of onset**

The time phase when the initial symptoms of the disease appeared, and when the child was examined and diagnosed will influence the understanding of CPI, the treatment process and other disease related features and consequences that are perceived differently due to different developmental stages. The psychological reactions to treatment and hospitalization of children varies in different ages (Hagglof, 1999). Psychological vulnerability in the early developmental stage can cause less resilience and reduce the ability to cope with the future developmental tasks.

**Disease duration**

Disease duration gives us a perspective on a developmental time phase of an illness. It is a meaningful dimension that completes the understanding of the psychological features of the chronic disorders. Each phase of the CPI has its own unique psychosocial demands and developmental tasks, which require significantly different strengths, attitudes, or changes from a patient and a family. Chronic disease time line and phases of illness were diagrammed by Rolland (Figure 4) (Rolland, 1994).

![Figure 4](image-url) Time line and Phases of illness (J.S. Rolland, 1994).

According to our objectives we were interested in a chronic phase of the diseases, the period that can be marked by constancy, progression, or episodic change; a psychosocial construct, that can be referred to as “the long
haul” or the phase of living day to day with chronic illness (Rolland, 1994). In this phase adolescents with CPI already have overcome the first stress of diagnosis and adjustment to treatment and new routine. At this stage adolescents and their families in different ways have adapted to the living with an illness.

**HbA1c**

HbA1c (diabetes group) – glycosylated hemoglobin is a form of hemoglobin mainly used for monitoring a long-term control of type 1 diabetes. Higher amount of HbA1c indicate poorer control of blood glucose level over the previous 2-3 months prior to the measurement. Metabolic control expressed in HbA1c is the most common indicator of disease severity in diabetic patients, and it has been shown to predict HRQoL in adolescents with diabetes (Hoey et al., 2001).
2. GENERAL OBJECTIVE

To assess the psychological well-being and family functioning in adolescents with chronic physical illnesses (CPI) in Northern Russia, as well as to identify factors of potential risk or protective significance for adolescents’ well-being.

2.1 Specific objectives

- To investigate the impact of chronic disorder (diabetes, asthma and epilepsy) on adolescents mental health and health-related quality of life;
- To study the effects of CPI on the family;
- To identify medical and socio-demographic factors associated with psychological well-being in adolescents with CPI.
3. SETTING

This thesis is based on four cross-sectional surveys. All studies were conducted between 2002 and 2003 in the city of Arkhangelsk, Russia. A brief description of the area, health care service system and national school system is justified to better understand the context where the studies have been conducted.

3.1 The city of Arkhangelsk

The study was conducted in the European Northern part of Russia – Arkhangelsk. The maps show the location of Arkhangelsk and studied area (Figure 5, 6).

![Figure 5. Location of Arkhangelsk (www.maps.google.com).](image)

Arkhangelsk is the capital of the region, with the population of about 375,000 at the time of the data collection. Arkhangelsk is divided into 8 administrative districts.

Arkhangelsk region by population density, climate and industry range represents a typical North European setting. However, given the drastic changes which happened in Russia since the fall of the Soviet Union, the region suffered some particular problems, though not unique in post-Soviet Russia. Mainly, these include a decrease in life expectancy, lower birth rate, ageing population, fall in industrial production and employment, increase of
alcohol-related problems and, last but not least, pessimistic economic and socio-demographic prognosis (Federal State Statistics Service 2014). The chosen sampling site represents an average Russian city, both by size, state of economy, social services, and socio-demographic situation. Therefore, I believe that our findings are, with a degree of caution, relevant for the whole Russia, and, probably, for some other post-Soviet states.

Figure 6. Map of Arkhangelsk.
3.2 Health care service

Public pediatric healthcare in Russia has a very long history. Dated back to 1763 it celebrated its 250 anniversary in 2013. The child health protection was established by the Empress Catherine II who signed the Manifest for foundation of Imperial Moscow Foster Home that much later grew into the Russian Academy of Medical Science. In the very first decades of the Soviet era a promising and effective system of child health care was created. It was successfully functioning until the collapse of the Soviet Union followed by a severe economic crisis in 1991. The political and economic changes since the late 80’s have profoundly affected all aspects of life in Russia. The health of the population, which had stagnated since the mid-1960s, has declined sharply since the late 1980s (Tkatchenko, McKee, & Tsouros, 2000).

The health situation in Russia during the 1990s was catastrophic. The federal spending on health care, the staple of the Soviet Union system, has continued to decline to bare minimal levels. Social and economic conditions are perhaps the largest contributors to the deterioration of population health, while the health leadership has wasted a decade with ineffectual and probably harmful, reforms that have failed to address the situation adequately (Varavikova, 2002).

For Russia the development of clear government health policy is a painful problem. The difficult situation in health care in 1990’s and beginning of 2000’s relates not only to the period of economic transition, but goes deep into the former Soviet health system. This state-operated service provided free, universal health care with sufficient, sometimes excessive resources in medical personnel, hospital beds, outpatient clinics, and other services. One may argue that in Soviet health care quantity was compromising quality. The system operated as a state monopoly, with the central government controlling budgets, setting mandatory norms, and rigidly controlling manpower training and research. It lacked mechanisms for epidemiologic or economic analysis, and accountability to the public (Varavikova, 2002). There has been no major health care reform since the fall of the Soviet Union until 2006, when the health care became one of the national priority projects.

Due to political and socio-economic factors state healthcare system is still predominantly operating in Russian Federation, although private system takes an average of 40% of services in some areas of medical practice. According to the law the state budget guarantees the coverage of all medical services, medication expenses and rehabilitation facilities for children with CPI. Never the less the commercialization of health care shifts significant share of the costs on the family, while many families are limited in resources to use private health services and treatments, physical education and sports.
3.3 National school system

School education in Russia is provided predominantly by the state. Education is compulsory up to the 9th grade. The stages of compulsory schooling are: primary education for ages 6-7 to 9-10; and secondary school for ages 10-11 to 14-15. If a pupil of secondary school wishes to get to the university, he or she must continue the school for two more years. Primary and secondary schools together comprise 11 years of study. Every school has a "core curriculum" of academic subjects. With the start of the secondary school children have multiple subject teachers, but there always is one class teacher, or a tutor (typically a subject teacher) for every group of children moving with his or her class throughout the years of education up until the end of the high school.
4. STUDY DESIGN, PARTICIPANTS, AND PROCEDURE

All the studies were cross-sectional. The analysis was performed between and within the groups. This thesis is based on adolescents’ sample of 148 adolescents with CPI and a reference group of 301 healthy adolescents’ self-ratings, parents’ ratings, and the evaluation of doctors in charge of the children with CPI (Table 1).

Table 1. Participants in CPI and Control groups.

<table>
<thead>
<tr>
<th>Sample type</th>
<th>Number, n</th>
<th>Study (n if different)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Diabetes group adolescents</td>
<td>50</td>
<td>X X X X X X</td>
</tr>
<tr>
<td>Diabetes group mothers</td>
<td>50</td>
<td>X X</td>
</tr>
<tr>
<td>Asthma group adolescents</td>
<td>50</td>
<td>X X X X (49) X</td>
</tr>
<tr>
<td>Asthma group mothers</td>
<td>50</td>
<td>X X</td>
</tr>
<tr>
<td>Epilepsy group adolescents</td>
<td>48</td>
<td>X X X X (47) X</td>
</tr>
<tr>
<td>Epilepsy group mothers</td>
<td>48</td>
<td>X X</td>
</tr>
<tr>
<td>Control group adolescents</td>
<td>301</td>
<td>X X X</td>
</tr>
<tr>
<td>Control group mothers</td>
<td>301</td>
<td>X X</td>
</tr>
</tbody>
</table>

4.1 CPI group

A total of 173 adolescents (90 girls and 83 boys) identified from outpatient clinic records, being treated and controlled by pediatricians were selected as potential participants. All adolescents with type 1 diabetes in Arkhangelsk who were referred to a child outpatient clinic at the start of the data collection period and who met our inclusion criteria (Table 2) were asked to participate in our study.

Table 2. Inclusion and exclusion criteria for the study sample.

<table>
<thead>
<tr>
<th>Participants CPI group</th>
</tr>
</thead>
<tbody>
<tr>
<td>Inclusion criteria</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Exclusion criteria</td>
</tr>
<tr>
<td></td>
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<tr>
<td></td>
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<tr>
<td></td>
</tr>
</tbody>
</table>
Altogether, 55 adolescents with type 1 diabetes matched our inclusion criteria. For the purpose of our investigation, we decided to include approximately the same numbers of adolescents with asthma and epilepsy. The subspecialists (endocrinologist, allergists, and neurologists) working in the pediatric outpatient clinic assisted in selection of the study sample. Studying the outpatient clinic registers from the three largest districts of Arkhangelsk, we chose for recruitment every second adolescent with epilepsy (n=59) and every third with asthma (n=59) matching our criteria (Table 2). Of the mothers who were contacted 91% agreed to participate in the study; refusal rates were higher in the epilepsy group – 15%. The adolescents who were inpatients during the period of data collection (n=5) were excluded from the study sample. Three families (2 with adolescents with epilepsy and 1 with asthma) were out of reach. Thus, the final sample consisted of 148 adolescents (diabetes (n=50); asthma (n=50), and epilepsy (n=48)) and their mothers. Mean adolescents’ age was 14.46. Socio-demographic and disease related characteristics of the study sample are presented in Table 3.

After obtaining consent from the outpatient clinic staff and the identification of the study population we contacted families with adolescents with CPI by phone. Those who agreed to participate in the study were asked to come to the outpatient clinic for the study meeting; several meetings were arranged to meet the preferred time for the participants. The author gave the overview of the study, assured about confidentiality of the reports, and explained the crux of the matter if the questions or statements seemed contradictory to mothers and adolescents. Thus, the test batteries were individually completed by CPI adolescents and their mothers. Those adolescent/mother dyads who didn’t find it possible to come to the outpatient clinic were contacted and the author visited them at home. Therefore, about half of the CPI participants were visited at their households. In both settings, the investigator observed the completion of the questionnaire to minimize the bias of parent/child presence.

4.2 Control group

Comparative data were obtained from a group of 301 schoolchildren and their mothers from secondary schools in each of three biggest administrative districts of the city. The first school in each of three districts where the informed consent was received from the principal was included in the study. One of three classes in each class parallel (from 7th to 10th grade) was selected. Thus our study included three schools, 12 classes (3 in each grade). Adolescents were chosen on the basis of age to match our subjects (Mean age 14.64). Any child with documented chronic illness, as noted in the school medical records, was excluded from the study. There were no drop outs in the control group. Control group adolescents filled the questionnaires at
school during regular class hours reserved for the study. The control group mothers received the test batteries from the class teachers during the parental meetings and were asked to complete it at home.

**Table 3.** Some socio-demographic and disease related characteristics of the study sample.

<table>
<thead>
<tr>
<th></th>
<th>Diabetes</th>
<th>Asthma</th>
<th>Epilepsy</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender Male/Female n (%)</td>
<td>28(56)/22(44)</td>
<td>27(54)/23(46)</td>
<td>19(39.6)/29(60.4)</td>
<td>131(43.5)/170(56.5)</td>
</tr>
<tr>
<td>Mean age</td>
<td>14.15</td>
<td>14.29</td>
<td>14.97</td>
<td>14.64</td>
</tr>
<tr>
<td>Parental age, mean</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother/Father</td>
<td>39.9/42.04</td>
<td>39.1/40.58</td>
<td>38.79/40.77</td>
<td>40.47/41.79</td>
</tr>
<tr>
<td>Changes in family (separation/death), n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>13 (26)</td>
<td>9 (18)</td>
<td>15 (31.3)</td>
<td>61 (20.3)</td>
</tr>
<tr>
<td>No</td>
<td>37 (74)</td>
<td>41 (82)</td>
<td>33 (68.8)</td>
<td>238 (79.1)</td>
</tr>
<tr>
<td>Missing</td>
<td></td>
<td></td>
<td></td>
<td>2 (0.6)</td>
</tr>
<tr>
<td>Family type, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Two-parent families (incl. step-father)</td>
<td>38 (76)</td>
<td>38 (76)</td>
<td>31 (64.6)</td>
<td>215 (71.4)</td>
</tr>
<tr>
<td>Single-mother family</td>
<td>12 (24)</td>
<td>12 (24)</td>
<td>17 (35.4)</td>
<td>86 (28.6)</td>
</tr>
<tr>
<td>Family SES, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>9 (18)</td>
<td>12 (24)</td>
<td>15 (31.3)</td>
<td>58 (19.3)</td>
</tr>
<tr>
<td>Average</td>
<td>28 (56)</td>
<td>18 (36)</td>
<td>18 (37.5)</td>
<td>122 (40.5)</td>
</tr>
<tr>
<td>High</td>
<td>12 (24)</td>
<td>16 (32)</td>
<td>14 (29.2)</td>
<td>98 (32.6)</td>
</tr>
<tr>
<td>Missing</td>
<td>1 (2.0)</td>
<td>4 (8)</td>
<td>1 (2.1)</td>
<td>23 (7.6)</td>
</tr>
<tr>
<td>Family size, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>3 (6)</td>
<td>3 (6)</td>
<td>7 (14.6)</td>
<td>27 (9)</td>
</tr>
<tr>
<td>3</td>
<td>22 (44)</td>
<td>25 (50)</td>
<td>17 (35.4)</td>
<td>101 (33.6)</td>
</tr>
<tr>
<td>4</td>
<td>17 (34)</td>
<td>21 (42)</td>
<td>16 (33.3)</td>
<td>131 (43.5)</td>
</tr>
<tr>
<td>5+</td>
<td>8 (16)</td>
<td>1 (2)</td>
<td>8 (16.7)</td>
<td>37 (12.3)</td>
</tr>
<tr>
<td>Disease severity, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Non-severe CPI</td>
<td>29 (58)</td>
<td>25 (50)</td>
<td>35 (72.9)</td>
<td></td>
</tr>
<tr>
<td>Severe CPI</td>
<td>21 (42)</td>
<td>25 (50)</td>
<td>13 (27.1)</td>
<td></td>
</tr>
<tr>
<td>Disease duration, mean (25th-75th centiles)</td>
<td>4.0 (3.0-6.0)</td>
<td>8.9 (6.6-10.9)</td>
<td>6.4 (3.2-9.7)</td>
<td></td>
</tr>
</tbody>
</table>

In Studies I, II, III, and IV all instruments were originally developed in English language. For Paper I and II the translation of the CBCL/YSR, BDI, and SFI questionnaires already existed. For Paper III and IV the translation of RSES and PAQLQ existed. The translation of “I think I am” self-evaluation scale, DQOLY and QOLIE-AD-48 into Russian was carried out following international recommendations on translation of a foreign instrument in a
new culture/society (Sartorius & Kuyken, 1994). A professional translator made a forward translation of the original version. The project executor raised some queries within the research group, the answers to which resulted in a forward translation. Another professional translator performed back-translation of the forward version into English. The comparison of the translation with the original was performed. The second Russian version of the questionnaires was developed with the emphasis on content validity.

The doctors in charge of the CPI patients – pediatricians with the specialization in a respective field endocrinology, allergology/immunology, or neurology evaluated the severity of the patients’ chronic condition in a paper-pencil format.
5. METHODS

5.1 Instruments

Child Behaviour Check-List (CBCL) and Youth Self Report (YSR)

The instruments consisting of 113/112 statements on a 0-2 scale, categorized in three main domains – Total problems, Internalizing, and Externalizing (Achenbach, 1991a, 1991b). In these instruments the respondents are asked to base their ratings on the preceding 6 months. The latest version of these questionnaires are the part of The Achenbach System of Empirically Based Assessment comprising an integrated set of forms for assessing competence, adaptive functioning, and problems (Achenbach & Rescorla, 2001). In our study we used two of three parallel forms developed by Achenbach to provide complete and comprehensive assessment of adolescents’ functioning. For our study analysis we used only Syndrome scales designated as Anxious/Depressed, Withdrawn scale, Somatic Complaints, Social Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior. The title of each summarizes the kinds of problems that form the syndrome. By “syndrome” Achenbach means “the set of concurrent things” but not the disease. These syndrome scores proved to be an excellent basis for comparing problems in different groups.

Previous research have shown that CBCL/YSR are valid and reliable instruments for assessing children and adolescents psychopathological symptoms, and this is also true for Russian versions of the scale (Carter, Grigorenko, & Pauls, 1995; Ruchkin, Eisemann, & Cloninger, 1998; Slobodskaya, 1999). Because the validation and adaptation to Russian of CBCL/YSR predated 2001 revision we used the 1991 version of the questionnaires in our study.

Beck Depression Inventory (BDI)

The BDI is a 21-item inventory of attitudes and symptoms (Beck, 1967; Beck & Steer, 1993). Statements are ranked to reflect the range of severity of depression from neutral to maximum severity. Numerical weighted values from 0 to 3 are assigned to each statement to indicate the absence of depressive symptoms (value = 0) or severe depressive symptoms (value = 3). Respondents are asked to describe themselves for the past week. The depression score on the BDI is the sum of the weighted response in all items. The possible range of scores is 0–63, with a score of 10–19 indicating mild to moderate; 19–29 moderate to severe; and above 30 severe depression. We
used the BDI-IA version of the questionnaire, the BDI-II version was not yet validated when the study was designed. Both versions are comparable in estimating being diagnosed with DSM-IV mood disorder (Beck, Steer, Ball, & Ranieri, 1996). A comparative validation study conducted on a Russian adult population noted that BDI could be recommended for screening diagnostics of depression, and reported the optimal cut off score for delimitation depressive patients: ≥12 (Andriushchenko, Drobizhev, & Dobrovol’skii, 2003). BDI has been widely used in adolescent populations both in psychiatric practice (Blom, Larsson, Serlachius, & Ingvar, 2010), and epidemiological studies screening for the depressive symptoms in a large school population (Olsson & von Knorring, 1999) showing acceptable psychometric properties in both settings.

**Self-report family inventory (SFI)**

The instrument consists of 36-item scores 1 to 5 and measures 5 family domains: Health/Competence, Conflict, Cohesion, Leadership, and Emotional Expressiveness (Beavers & Hampson, 1990). The instrument may be completed by family members 11 years of age and older. The respondents are requested to estimate to what extent the statement fit into their family situation. The scale descriptors are 1 = Yes: Fits our family very well, 3 = Some: Fits our family some, and 5 = No: Does not fit our family. The last two items have response scales specific to the items. Lower scores represent greater family competence. This questionnaire is based on the Beavers System Model (Beavers & Hampson, 2000). The model suggests that optimally functioning families differ from less capable or competent families with regard to dimensions of family structure (characteristics of leadership, parental coalitions and family members’ closeness to each other), mythology (how the family view itself), goal-directed negotiation, development of autonomy in individual family members and the nature of family affect and affective expression. SFI gives a possibility to capture several domains from the numerous specific family variables: competence, which defines how well the family performs the necessary and nurturing tasks of organizing and managing itself; cohesion, estimating family style which addresses closeness, togetherness and tendencies to enjoy time and activities together; and clinically useful scales of conflict, leadership and emotional expressiveness. Psychometric evidence of SFI reliability and validity is substantial; studies demonstrate a 91% correct classification of clinical versus non-clinical cases, high test–retest reliability, high internal consistency and concurrent validity (Halvorsen, 1991). This questionnaire was translated into Russian language, used and validated on a special subgroup of male adolescents (n =159) by Ruchkin et al. (Ruchkin, Koposov, & Hagglof, 2000).
Quality of Life in Epilepsy Inventory for Adolescents (QOLIE-AD-48)

The instrument has been developed and validated to assess multiple aspects of HRQoL in adolescents with epilepsy (Cramer et al., 1999), it contains 48 items in 8 sub-scales: Epilepsy Impact (12 items), Memory/Concentration (10 items), Attitude toward Epilepsy (4 items), Physical Functioning (5 items), Stigma (6 items), Social Support (4 items), School Behaviour (4 items), Health Perception (3 items), and a Total summary score. The raw numbers of the five-point Likert scale for QOLIE-AD-48 domain scores can be converted into a 0-100 point response scale, with higher scores indicating better HRQoL. QOLIE-AD-48 has been shown to be a specific, sensitive and reliable measure and has been translated and validated for Chinese, Serbian and Brazilian populations (Barbosa, Guerreiro, & de Souza, 2008; Stevanovic, Lozanco-Miladinovic, Jovic, & Sarenac, 2005; Wang, Wu, Zheng, Zhang, & Li, 2009). The developer (Ms. Joyce Cramer) provided the original QOLIE-AD-48 version.

The Diabetes Quality of Life Questionnaire for Youths (DQOLY)

The instrument, developed by the Diabetes Control and Complication Trial Research Group (The DCCT Research Group, 1988) and later modified by Ingersoll and Marrero (Ingersoll & Marrero, 1991), is composed of 52 items in four subscales: a Diabetes Life Satisfaction scale, a Diabetes Impact scale, a Diabetes-Related Worries scale, and a single item general self-rating of overall health in a response format of a four- or five-point Likert scale. In all subscales, except for the Life Satisfaction subscale, lower scores indicated higher HRQoL. The scales can be recorded in the same direction for ease of cross-scale comparison. The Hvidøre Study Group of Childhood Diabetes translated and used DQOLY in many countries. The questionnaire proved to be a reliable source of information on adolescent HRQoL, and a comprehensive and workable tool (Hoey et al., 2001).

Pediatric Asthma Quality of Life Questionnaire (PAQLQ)

The PAQLQ is a 28-item questionnaire designed for children and adolescents with asthma (Juniper et al., 1996; Juniper, Guyatt, Willan, & Griffith, 1994). It was derived from a HRQoL questionnaire for adults with asthma (Juniper et al., 1992). The child version uses a seven-point Likert scale that can be completed reliably by children as young as seven years. The PAQLQ includes a total score, and three primary domains: how asthma interferes with activities, asthma symptoms, and emotional reactions to asthma. Higher scores in these domains indicate better HRQoL. PAQLQ has been shown to have good measurement properties and high validity in both evaluative and discriminative studies (Juniper et al., 1996). Translation into
Russian and linguistic validation of PAQLQ was made by the MAPI Research Institute in Lyon, France (MAPI Research Institute).

**The Rosenberg Self-Esteem Scale (RSES)**

The instrument is a 10-item Guttman scale of self-esteem (Rosenberg, 1965). Respondents are asked to strongly agree, agree, disagree, or strongly disagree with each of the items. “Positive” and “negative” items are alternated in an attempt to reduce the effect of respondent bias. At least six items must be scored as positive for a respondent to be considered to have low self-esteem. This instrument is one of the most widely used for evaluating the global feeling of self-worth. It was validated extensively for high school students and is the most accepted self-esteem scale in this population (Robins, Trzesniewski, Tracy, Gosling, & Potter, 2002). RSES have been used earlier in the research of adolescents with CPI (Seigel, Golden, Gough, Lashley, & Sacker, 1990). This questionnaire was translated into Russian language, and used on a special subgroup of male adolescents versus controls by Ruchkin (Ruchkin, Eisemann, & Hägglöf, 1999).

**I think I am**

The instrument is the measure of self-evaluation consisting of 72 statements scored -2 to +2 (Ouvinen-Birgerstam, 1985). The questionnaire can be used in children 10 to 18 years of age. The complete scale can be divided into five sub-scales: “Physical characteristics”, “Skills/talents”, “Psychological well-being”, “Relationship to the family” and “Relation to others”. Age and gender are taken into consideration before the final score is expressed in the standardized nine-point stanine scale. Higher scores indicate higher self-esteem. This instrument was standardized on Swedish children and commonly used in medical research studying self-esteem of children with different CPI (Wennstrom, Berg, Kornfalt, & Ryden, 2005).

**Clinical data**

Clinical data were drawn from the medical records of the patients at the outpatient clinics. These included the type of disease, age at clinical diagnosis, disease duration and, for adolescents with diabetes, HbA1c levels. According to medical records the mean age of the disease onset in patients was as follows: diabetes 8.9 ± 3.3; asthma 5.8 ± 2.8; epilepsy 8.3 ± 3.4. The median disease duration in adolescents with CPI is presented in Table 3. The median HbA1c level was 10.42% (interquartile range 7.3 - 12.1%). The median number of daily insulin injections was five (interquartile range 5–6). Disease severity was evaluated by doctors in charge of the cases. Two questions (one about the level of the disease control and one about the patient’s current condition during the recent follow-up visit) each with four
answer alternatives ("very poor" = 1, "poor" = 2, "good" = 3, and "very good" = 4) were asked in paper-pencil format. Physicians defined acceptable disease control based on the following determinants: (a) epilepsy – type of seizure, seizure frequency, antiepileptic medications and observed side effects, number of hospitalizations; (b) asthma – frequency of asthmatic episodes, medication side effects, number of hospitalizations; and (c) diabetes – level of metabolic control, number of hypoglycaemic episodes, disease complications, and number of hospitalizations. These scores were summed. Total score of ≥6 were designated “non-severe”, and scores of ≤5 coded “severe”. The score of 5 was selected for the severe disease status because it required at least one of the categories to be evaluated as “poor” or “very poor”.

**Socio-demographic status**

Since no SES classification scheme was available for Russia, we used the mother’s/father’s education level, occupation level, and income level. For education, completed high school scored 0, college education and incomplete university education scored 1, and five or more years of university scored 2. For occupation, unskilled manual workers scored 0, skilled manual workers scored 1, and white-collar workers scored 2. For income level, there were three alternatives: low scored 0, average scored 1, and high income scored 2. The income level score was reached by the grouping of data, dividing the study population reports into 3 groups, representing each low, average, or high 33% of the data.

In order to analyze the relationship between psychological aspects and socioeconomic variables we performed certain recoding manipulations. In two-parent families, the highest rating for parent occupation and education was used. The categories were then summed into the following classes, where 2 = high family SES (range 5-6), 1 = average SES (3-4), and 0 = low SES (0-2).

**Family related factors**

Family type (a parameter reflecting two- or single- parent household), family size, and changes in the family life (parent separation/death) were included into the study analysis.
5.2 Statistics

Continuous data were described using mean with standard deviation (SD) or median with 25th - 75th centiles. Cronbach’s $\alpha$ was used to determine internal consistency reliability of the questionnaires. Differences between independent groups for continuous data were quantified with Independent – Samples t-test in case of observation approximating the normal distribution and with Mann-Whitney test in case of the severe skewness of observations or too few observations. For categorical data Fisher’s exact test was applied. Differences in mean between related samples were analyzed with Paired-Sample t-test. Correlations were analyzed with Pearson’s correlation. General linear model (GLM) was used to investigate the predictive value of explanatory variables and selected outcomes. The effect size was estimated by partial $\eta^2$ measure. Path analysis was used to describe dependences between the set of variables. Path analysis is a kind of multivariate analysis in which causal relations among several variables are represented by graphs (path diagrams) showing the “paths” along with each causal influences travel. Path analysis can be viewed as a case of structural equation modeling (SEM), when only single indicators are employed for each of the variables in the causal model. An advantage of path analysis is that it allows for the calculation of direct and indirect affect of independent variables.

The Statistical Package for the Social Sciences (SPSS 22) and an add-on module Analysis of Moment Structure (AMOS) was used for the analyses. SEM was used to evaluate the mediating effects of the variables specified by models with path diagrams. This software performs analyses of moment structures through maximum likelihood estimation to investigate whether a variable X (Family functioning) is a mediator between independent variable A1 (Adolescent and Family demographics)/A2 (Disease severity) and dependent variable B1 (Mental Health) and B2 (HRQoL) in the path analysis. When testing the factor loading p-value <.05 was considered significant. The effects are expressed in standardized regression coefficients. When evaluating the path model the following criteria were used: CMIN/DF – the minimum discrepancy divided by its degrees of freedom, smaller values preferable; GFI – goodness of fit index, a value of 1 indicates a perfect fit; AGFI – adjusted goodness of fit index is bound above by one, which indicates a perfect fit; CFI – a comparative fit index, a values close to one indicate a very good fit, RMSEA – a root mean square error of approximation, smaller values, below .08 preferable.

Missing values in the data set were replaced by the average scores for the pertinent items. Though, there were very few missing values in the data set.
5.3 Ethics

There was no ethical research committee in Arkhangelsk at the time of project planning and data collection. Informed consent was obtained from the head of the respective child outpatient clinic and school principals. All participants were informed that participation in the study was voluntary and confidential. Informed consent was obtained from all the participants (both mothers and adolescents). Participants were assured that outpatient clinic staff would not receive any private information from the questionnaires.
6. RESULTS

6.1 Mental health in adolescents with CPI (Paper I and III)

Overall the results obtained showed certain differences in mental health of CPI adolescents versus healthy counterparts, although there were discrepancies in the mental health pattern depending on the informants. There were no significant differences found in self-reported Total behavior-emotional problems and Depressive symptoms of CPI adolescents compared to the controls. This study found that parents endorse more behavioral problems than CPI children. Considering mother’s reports alone, it is confirmed that adolescents with CPI have noticeably more behavior-emotional problems. In our study sample, internalizing problems were more pronounced within the index group, which is a common occurrence among children with CPI even if they do not reach clinical levels (Pinquart & Shen, 2011b). On the other hand, in our study adolescents with CPI reported much less externalizing-type problems compared to controls. Our findings distinguished adolescents with diabetes as having fairly good mental health state and not being at risk for behavior-emotional problems compared to asthma and epilepsy group peers. The most pronounced behavior-emotional problems and depressive symptoms were found in adolescents with asthma and epilepsy. Disease severity played a crucial role in behavior-emotional status and presence of depressive symptoms. The presence of behavior-emotional problems in asthma and epilepsy group were related to gender. Furthermore, elevated risks for developing behavior-emotional problems were found in single-parent households and in families with low SES.

Self-esteem as another dimension of mental health is presented in Paper III. Comparative analysis of the whole CPI group versus Controls showed significant differences between the self-esteem reports. Unexpectedly adolescents with CPI reported better psychological well-being and higher self-esteem (RSES).

Adolescents with diabetes scored higher on Psychological well-being, Relations to family, Total “I think I am” scale, and RSES than controls. While adolescents with epilepsy scored lower on Skills, Relations to family, and Total “I think I am” scale than control group. No differences were found between adolescents with asthma and controls.
Within the CPI group adolescents with diabetes reported generally higher self-esteem than their peers with asthma and epilepsy. While adolescents with epilepsy showed lower self-esteem than those with asthma.

General linear model was used to analyze the predictive value of certain demographic (adolescent age, gender, SES), family (family size, single-parent household, parental separation/death), and disease-related factors (disease severity, disease duration and age of onset, as well as HbA\textsubscript{1c} in a diabetes group) on adolescent self-esteem. The analysis revealed diverse factors contributing self-esteem perception in three CPI groups. Disease severity and SES were associated with self-esteem in diabetes and asthma groups, while age and gender related to self-esteem in adolescents with epilepsy.

### 6.2 Health-related quality of life in adolescents with CPI (Study IV)

The internal consistency reliability was found to be satisfactory for all DQOLY subscales. Transformed subscales scores were below 50%, indicating good HRQoL. Adolescents with diabetes expressed most concerns regarding the impact of the disease and life satisfaction. Among this study group 52% evaluated their overall health as “Fair”, 36% “Good”, 8% “Excellent”, and 4% “Poor”. Girls with diabetes showed significantly more disease related worries than boys.

The internal consistency reliability, assessed with the Cronbach alpha coefficient, was satisfactory, with all PAQLQ subscales above the conventional standard of $\geq 0.70$. The possible total score range was from 1 (maximum impairment) to 7 (no impairment); the sample range was from 3.35 to 7. The mean Overall score was 5.7, SD 0.92. Adolescents with asthma estimated their HRQoL toward positive end of each specific scale. They expressed most concerns in Activity limitations domain. Girls with asthma reported more impairment in HRQoL compared to boys, they scored significantly lower on the Symptom domain and Overall score.

Internal consistency of QOLIE-AD, assessed by Cronbach’s alpha coefficient, was found to be satisfactory ($\geq 0.70$) only in the Memory/concentration, Attitude, Stigma and Total subscales. The Total mean score for QOLIE-AD was also toward the positive end of the scale. The most considerations of patients with epilepsy were expressed in two subscales – Attitudes toward epilepsy and Stigma.

Multiple linear regression was used to analyze the predictive value of certain demographic, family, and disease related factors on HRQoL. Disease severity predicted HRQoL outcome in diabetic group and to some extent in asthma.
group. For all three groups gender had an evident impact. In our sample socioeconomic status predicted HRQoL in patients with asthma and diabetes. Age was a determinant of the HRQoL level in adolescents with epilepsy.

6.3 Family functioning in adolescents with CPI (Paper II)

This study examined the association of childhood chronic physical illnesses with family functioning by comparing to healthy peer families. Comparing to our hypothesis there were no differences found between these two groups in both mother and adolescent reports.

The perception of family functioning differed a lot between CPI groups. Diabetes adolescent/mother reports showed considerably more functional families than the reports from asthma and epilepsy group, they scored higher Health/Competence, less Conflict, higher Cohesion and higher Emotional Expressiveness. Mothers of adolescents with epilepsy reported lower Conflict and higher Family Cohesion compared to mothers from asthma group.

Family functioning was significantly associated with SES and family type both in CPI and control group; and also with disease severity and disease duration in CPI group. Greater adolescent disease severity was associated with living in a more dysfunctional family, characterized by lower levels of system competence, poor emotional expressiveness and rigidity. Unexpectedly, control group adolescents perceived their families as less functional compared to patients in non-severe state of the disease, they reported less competence and lower family cohesion.

Paired-sample t-test used for the whole study sample (patients and controls) revealed discrepancy in the perception of family functioning between the informants. There was a significant difference between mothers’ and adolescents’ ratings in all aspects of family functioning but conflict. Adolescents perceived their families as less competent, less cohesive and less emotionally expressive compared to their mothers.

6.4 Factors of potential risk and protective significance for psychological well-being in adolescents with CPI

The tentative model

The main aim of the present analysis was to develop and test a model on psychological well-being on data from adolescents with three different CPI
and their mothers with potential possible factors involved in youths’ mental health state and self-perceived HRQoL with the starting assumption about the connection between mental health/HRQoL and family functioning. Table 4 summarizes the factors included in the tentative model.

**Table 4. Tentative conceptual model on psychological well-being of adolescents with CPI.**

<table>
<thead>
<tr>
<th>Antecedent factors</th>
<th>Observed exogenous factors</th>
<th>Observed endogenous factors</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Disease severity</td>
<td>- Pediatrician evaluation</td>
<td>Family functioning</td>
<td>Mental Health</td>
</tr>
<tr>
<td></td>
<td></td>
<td>- Health/Competence</td>
<td>YSR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Adolescent perspective</td>
<td>- Total Problems YSR</td>
</tr>
<tr>
<td>Disease duration</td>
<td></td>
<td>- Health/Competence</td>
<td>CBCL</td>
</tr>
<tr>
<td>Age of onset</td>
<td></td>
<td>Adolescent perspective</td>
<td>- Total Problems</td>
</tr>
<tr>
<td>Adolescent demographics</td>
<td>Age</td>
<td>Mother perspective</td>
<td>Depression</td>
</tr>
<tr>
<td></td>
<td>- Sex</td>
<td></td>
<td>- BDI Total score</td>
</tr>
<tr>
<td>Family demographics</td>
<td>- SES</td>
<td>Self-esteem</td>
<td>- RSES</td>
</tr>
<tr>
<td></td>
<td>- Changes in family</td>
<td>Self-concept</td>
<td>- I think I am Total score</td>
</tr>
<tr>
<td></td>
<td>(separation/death)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>- Family type (two/single parent household)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>- Family size</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

The exogenous variables such as adolescent- and parent- demographics, as well as the disease severity status are hypothesized to account for differences in family functioning, mental health and HRQoL. Three antecedent factors (disease duration, age of onset and family size) and one outcome factor “depression” were not included in the models due to the low Person’s correlation coefficients identified. Thus, six antecedent factors were included – child gender, child age, family SES, changes in family, family type, SES, and disease severity.

“Disease severity” was tested as an antecedent factor considering that mental health and HRQoL is influenced by disease severity state (Eiser, 1990).

The factor “HRQoL” was tested as a consequence in our disease-specific models. HRQoL is known as an outcome measure reflecting physical fitness, social integration and support, psychological stability and ability to fulfill daily tasks across ages and gender (Sartorius et al., 1993). There was no possibility to include this factor to the general CPI model, because only disease-specific questionnaires were used to study HRQoL.
The conceptual tentative model is specified as a path model. Different pathways that relate family functioning reported by mother/adolescent dyads to mental health and HRQoL were tested with measures including all antecedent factors. Before testing the models the intercorrelations among the measures were analyzed. The factor was included in the models if the path coefficient was significant. All regression coefficients signs were in the expected direction. Based on a theoretical view we evaluated a tentative conceptual model first using all CPI study sample; second, including each one of three CPI adolescents groups (diabetes, asthma, and epilepsy).

We were using path analysis to test whether our hypothesis about the relationships between variables are plausible, supported by the data from three different CPI groups, and represent underlying processes.

Path diagram for hypothetical model predicting self-reported mental health of adolescents with CPI (n=148) is presented in Figure 7.

**Figure 7.** Path diagram for hypothetical model predicting self-reported mental health in adolescents with CPI (Model 1).

Note: Path loadings are standardized coefficient CMIN/DF= 1.75; GFI= 0.95; AGFI= 0.89; RMSEA= 0.07
Model 1 suggests that the level of family functioning will mediate the relation between the three factors child age, disease severity, family type and the level of self-reported mental health.

**Figure 8.** Path diagram for hypothetical model predicting psychological well-being in adolescents with diabetes (Model 2).

Note: Path loadings are standardized coefficient CMIN/DF= 1.40; GFI= 0.85; AGFI= 0.73; RMSEA= 0.09

Model 2 based on data from adolescents with diabetes (n=50) and their mothers (n=50) is presented in Figure 8. The model revealed two demographic factors (child age and changes in the family) and disease severity indirectly influencing psychological health of adolescents. They were mediated by self-perceived family functioning level.

The model suggested mother’s perception of family functioning as the mediating factor between two interconnected background factors - changes in the family and single-parent household- and the level of behavior-emotional problems.

All mental health contributors but self-esteem failed to explain the variation in HRQoL.
Model 3 based on data from adolescents with asthma (n=50) and their mothers (n=50) is presented in Figure 9. The model provided support for the mediating role of self-perceived family functioning in which the antecedent factors – child gender, disease severity and family SES – are viewed as the determinants of mental health.

Two background parameters – family SES and family type – indirectly influenced mother’s reports on behavior-emotional problems of adolescents, being mediated by the level of family functioning.

All mental health contributors but self-esteem failed to explain the variation in HRQoL in asthma group.
Model 4 based on data from adolescents with epilepsy (n=48) and their mothers (n=48) is presented in Figure 10. The antecedent factors – child gender, family SES and family type – had significant pathways connecting to youth-perception of family functioning, which mediated self-evaluated mental health level.

The family functioning level perceived by mothers failed to mediate the relationship between the antecedent factors and mental health regardless the informant. Self-esteem and self-concept were the second order mediators of the HRQoL level perceived by adolescents with epilepsy.

Several goodness of fit measures were used and those are presented in Table 5. All models but epilepsy model (Model 4) satisfied the CMIN/DF criteria and are relatively close to the desired values level for GFI, AGFI and CFI. The exceptions are the values on RSMEA in asthma and epilepsy models (Model 3 and 4).
Table 5. Goodness of fit measures for Structural Equation Models.

<table>
<thead>
<tr>
<th></th>
<th>Model 1 CPI</th>
<th>Model 2 Diabetes</th>
<th>Model 3 Asthma</th>
<th>Model 4 Epilepsy</th>
</tr>
</thead>
<tbody>
<tr>
<td>CMIN/DF</td>
<td>1.75</td>
<td>1.40</td>
<td>1.84</td>
<td>3.09</td>
</tr>
<tr>
<td>GFI</td>
<td>0.95</td>
<td>0.85</td>
<td>0.81</td>
<td>0.80</td>
</tr>
<tr>
<td>AGFI</td>
<td>0.89</td>
<td>0.73</td>
<td>0.67</td>
<td>0.58</td>
</tr>
<tr>
<td>CFI</td>
<td>0.96</td>
<td>0.93</td>
<td>0.82</td>
<td>0.82</td>
</tr>
<tr>
<td>RMSEA</td>
<td>0.07</td>
<td>0.09</td>
<td>0.13</td>
<td>0.21</td>
</tr>
<tr>
<td>P value</td>
<td>0.03</td>
<td>0.05</td>
<td>0.00</td>
<td>0.00</td>
</tr>
</tbody>
</table>
7. DISCUSSION

The studies presented in this thesis explore behavior-emotional problems, depressive symptoms, self-esteem, health-related quality of life, and family functioning in a sample of adolescents with chronic illnesses. Comparisons are made with healthy counterparts. Furthermore, the factors of potential risk and protective significance contributing to the psychological health of adolescents with CPI are described and discussed.

Mental Health

The presented findings suggest that adolescents with CPI are at increased risk for developing internalizing problems, while healthy counterparts are more likely to externalize their problems. In 2011 published meta-analysis of the behavior problems in children and adolescents with CPI, Pinquart and Shen, found that the frequency of internalizing problems, social problems, attention problems and thought problems were higher than the frequency of externalizing problems (Pinquart & Shen, 2011b). The explanations of elevated internalizing problems in children with CPI were assumed in the latter study; they included the perceived lack of control over the illness and its symptoms or progression (e.g. epilepsy or sickle cell disease), frightening symptoms (e.g. in the case of asthma or epilepsy), restrictions in positive activities (e.g. due to hospitalization), peer rejection, as well as side effects of therapy. The risk factors for internalizing problems may be more widespread and/or may have a stronger effect on adolescents with CPI than risk factors of externalizing problems.

The statistically significant lower levels of externalizing problems in CPI adolescents compared to healthy counterparts may be explained by possibility that CPI children experience aggressive behavior as a dysfunctional coping strategy in their position. Since it could evoke physical aggressive reactions from others that they are not able to deal with (Meijer, Sinnema, Bijstra, Mellenbergh, & Wolters, 2000). Taking into consideration the homogeneity of the study sample and no SES differences the potential explanation of lower delinquency levels in CPI versus healthy population might be that CPI adolescents are trying to be socially desirable and practice appropriate social behavior. Another explanation might be in the family. Surrounded by a caring and protective social environment CPI children tend to display more submissive and less assertive behavior (Meijer et al., 2000). In accordance with this Cappelli et al. stated that parental overprotection plays an important role in psychological functioning of the CPI children (Cappelli et al., 1989). CPI might be considered as a protective factor taking into account mild disease severity, successful family functioning and parental rearing practice. Several studies have suggested the protective
effects of CPI on health-related behavior (Jacobson et al., 1997), but not on psychological well-being.

An interesting outcome was found in the cross informant analysis of mother/adolescent reports of CBCL/YSR questionnaires. Adolescents with CPI reported fewer behavior-emotional problems than their mothers, in contrast to healthy adolescents who scored significantly higher compared to mothers. The pattern of underreporting the symptoms in children with CPI was found earlier and explained by the possibility that children with CPI want to present themselves as healthy functioning individuals (Huberty, Austin, Harezlak, Dunn, & Ambrosius, 2000; Martin, Ford, Dyer-Friedman, Tang, & Huffman, 2004). Alternatively, parents might underestimate the child’s ability to adapt toward their illness (Pinquart & Shen, 2011b). Parents’ reports can also be biased by parental distress caused by child CPI. Based on the literature healthy youths themselves endorse more problem behaviors than did any of the evaluating adults, this pattern is a characteristic of all symptoms but withdrawn behavior (Grigorenko et al., 2010). Our results once again prove the importance of incorporation of information from multiple informants when mental health characteristics are evaluated in children and adolescents.

Another aspect of mental health that is in focus of this thesis is self-esteem in adolescents with CPI. DuBois and Flay reveal considerable support for the idea that achieving a high level of self-esteem, or perhaps more important, avoiding low self-esteem, is essential for health and well-being throughout the life span. High self-esteem has been included consistently among criteria used to define positive mental health (D.L. DuBois & Flay, 2004). A Swedish prospective population study found that positive self-esteem in childhood is a protective factor for mental health and quality of life reported in adulthood (Cederblad, 1996). The maintenance of positive self-esteem is believed to be important for psychological well-being, adjustment, and effective management of the chronic illness in adolescence (Helgeson, Snyder, Escobar, Siminerio, & Becker, 2007; Schneider et al., 2009). The findings of this thesis give support to that approach.

In our study adolescents with various CPI showed diverse levels of self-esteem. Adolescents with diabetes scored higher on all but one aspects of self-concept compared to their peers with epilepsy, and higher on a general level compared to adolescents with asthma. This was not an unexpected finding considering the previous results where adolescents with diabetes proved to show fairly good mental health state and not being at risk for developing behavior-emotional problems compared to their peers with asthma and epilepsy (Zashikhina & Hagglof, 2007). The self-esteem ratings in the asthma group were relatively high. Supporting the results of Austin et al. study where midadolescence asthma youths scored close to population norms and were not at risk for behavioral problems (Austin et al., 2000).
Comparison analysis of the three disease groups of our study revealed that adolescents with epilepsy reported lowest self-esteem. This outcome is in accordance with earlier research; Chiou and colleagues reported decreased levels of self-concept in children with epilepsy compared to their peers with asthma (Chiou & Hsieh, 2008). The reasons behind this outcome can be understood by means of the different dimensions of adolescent life: the CNS involving disease, low levels of family functioning (Austin et al., 2010), the lack of social support, and the stigmatization (discussed below).

Adolescents with diabetes reported higher self-esteem than their peers from the control group, and there were no significant differences between the reports of adolescent with asthma versus controls. Helgeson study also did not find any difference between adolescents with diabetes and control group regarding depressive symptoms, behavioral problems or self-worth over time (Helgeson et al., 2007). Generally, the comparison studies on self-esteem and self-concept fail to find differences in the reports of adolescents with diabetes versus controls (Vlachioti et al., 2010). Diabetes and asthma receive a major attention from all kinds of pediatric healthcare professionals. They help and support youths through the long period of awareness of disease after diagnosis; convince adolescents on compliance with follow-up visits and proper disease management; give education and knowledge on the disease itself and its course. All this gives a possibility for the youths to avoid hypersensitivity, instability, self-consciousness, and lack of self-confidence – chief characteristics of low self-esteem identified by Rosenberg and Owens (Rosenberg & Owens, 2001). Adolescents with epilepsy, in contrast, did receive lower attention and support from health care. Taking into consideration the stigmatizing effect of the disease involving CNS, the prejudice and social conventions around them it was expected to distinguish epilepsy group as most vulnerable.

**HRQoL**

In this thesis we were able to examine health-related quality of life in three cohorts of pediatric patients using disease-specific instruments, focusing on the self-perceptions of adolescents with diabetes, asthma or epilepsy. The outcomes in all three studied groups are quite reassuring. The scores of all DQOLY subscales – Diabetes Life Satisfaction, Diabetes Impact and Diabetes-Related Worries - indicated a relatively high level of HRQoL, although the single item indicating overall health of adolescents with diabetes was evaluated in a negative direction by 56% of participants. This discrepancy in our data would therefore suggest that the perception of HRQoL in diabetic adolescents is influenced not only by health related parameters but by the complex of non-disease related determinants. The maintaining of optimal HRQoL in children with type 1 diabetes was stated by Wagner et al., reporting similar levels of HRQoL in participants with
diabetes as in their healthy peers (Wagner, Muller-Godeffroy, von Sengbusch, Hager, & Thyen, 2005). In our sample, adolescents expressed less Disease-Related Worries, but lower Life Satisfaction compared to the original Ingersoll and Marrero study (Ingersoll & Marrero, 1991). Adolescents with asthma rated their HRQoL toward the positive end of each specific scale. The overall score of PAQLQ calculated from our study sample reports were relatively high and similar to the results described in other studies (Alvim et al., 2009; Annett, Bender, Lapidus, DuHamel, & Lincoln, 2001; Reichenberg & Broberg, 2000). Adolescents with asthma expressed most concerns in the Activity limitations domain. Asthma is a disease leading to a more restricted lifestyle; Williams described adolescents’ perceptions of these limitations (Williams, 2000). The most significant considerations of patients with epilepsy were expressed in two subscales – Attitudes toward epilepsy and Stigma. The same kinds of conclusions were drawn by Wang and colleagues in validation of the Chinese version of QOLIE-AD (Wang et al., 2009). Negative attitudes in society towards epilepsy can be the obvious and complex explanation of this finding, since stigma has an adverse impact on patients’ psychological wellbeing and HRQoL (Guekht et al., 2007). For many patients, stigma is a continuing social reality of their condition (Jacoby & Austin, 2007). This may lead to adolescents incorporating CPI into their individual personal and social identities in diverse ways, which can affect how they choose to live with the illness (Williams, 2000) as well as how they perceive their HRQoL.

**Family Functioning**

One of the most difficult of all human experiences is to accept a serious illness in the young; the profound sense of loss and unfairness inherent to this experience makes it one of the most challenging situations for families to master (Rolland, 1994). Opposite to hypothesis, families with adolescents with CPI showed considerable resilience and tolerance to the disease. There were no differences found between CPI versus control group in the measures of family functioning. This finding is consistent with other research focused on children with CPI and their families (Gerhardt et al., 2003; McClellan & Cohen, 2007; Reiter-Purtill et al., 2008). Overall, we support Rodrigues and Patterson explanation of the same type of finding by the possibility that the presence of a stressor like having a child with a CPI challenged these families to develop internal strengths for managing their situation, not taking into consideration other external factors as disease duration, disease severity and socio-economic variables (Rodrigues & Patterson, 2007). Rolland argues that the actual impact of CPI on the family depends on the psychosocial effect of illness and the pre-illness characteristics of the family, reflected in culturally defined beliefs (Rolland, 1994). As it was mentioned above, diabetes, asthma and epilepsy are nonincapacitating chronic conditions according to Rolland and Walsh classification (Rolland & Walsh, 2006). We
can assume that cultural features and beliefs (e.g. rigid gender-defined roles in the family, stigma associated with the illness) do not interfere with the family unit and do not disrupt the family functioning. However, there were differences in family functioning between CPI groups. The most problematic functioning was reported by families with adolescents with asthma. This finding may be partly explained by characteristics of our study sample – the longest disease duration in adolescents with asthma. Nevertheless, this outcome is in line with the earlier research reporting disturbed family interaction with regard to adaptability and cohesion in families with children with asthma compared to controls or families with children with diabetes (Gustafsson, Kjellman, Ludvigsson, & Cederblad, 1987). In our study sample mothers and their children with diabetes perceived families as well-functioning compared to participants with asthma and epilepsy. We assume that the explanation of this finding is in characteristics of diabetes as a disease itself and its perception and acceptance in the society. First, the society has generally a positive attitude towards diabetes compared to beliefs about other CPI as asthma or epilepsy. Westbrook studying attitudes towards disabilities showed that diabetes and asthma are the most accepted diseases (Westbrook, Legge, & Pennay, 1993). Epilepsy however, as disabilities involving CNS is often considered a stigmatizing condition. Second, in the last few decades’ quality of medical services for patients with diabetes was constantly growing, therefore it has become easier to manage the illness and gain better disease control resulting in the healthier family outcome.

Factors of potential risk and protective significance for psychological health of adolescents with CPI

This analysis can be viewed in the context of the Wallander et al. (Wallander et al., 1989) risk-resistance model as an examination of the contribution of the socio-ecological resistance factors in studied population. In this study, measures of adolescent’s mental health and HRQoL are viewed as indices of “child adaptation”. The analysis is focused on the importance of successful family functioning in adolescents with CPI, and socio-demographic and disease related factors influencing psychological well-being. First, a hypothetical model was tested, that includes the factors assumed to be related to mental health of CPI adolescents. Only youth-self-reports on mental health and family functioning were included in the first model. The tested model confirmed the hypothesis about the connection between family functioning and mental health in CPI adolescents, in that the level of family functioning mediates the relationship between the observed exogenous antecedent factors and the level of mental health. Older child age, higher disease severity, and single-parent household were the risk factors that indirectly influenced mental health through the prism of family functioning in adolescents with CPI. Previous research has shown the importance of the
studied parameters on mental health outcome (Cederblad, Dahlin, Hagnell, & Hansson, 1994; Lewis & Vitulano, 2003; Perrin et al., 2012; Ruchkin, Koposov, & Hagglof, 2000). Disease severity had the biggest effect among the antecedent factors. This contradicts the findings of Rodrigues and Patterson in the study with a noncategorical approach to the chronic conditions who found very little of the variance in family functioning explained by severity of child’s illness (Rodrigues & Patterson, 2007). Family functioning had a larger effect on personality factors (self-esteem and self-concept) compared to behavior-emotional problems and depressive symptoms. Therefore, we can conclude that self-esteem/self-concept of CPI children is hampered by “poor family functioning” to higher degree than other mental health components. The basic capacity to feel good about oneself in adolescents with CPI is most vulnerable when contradicted by family dysfunction. Adolescence by itself presents some of the most intense parent-child confrontations around the issues of control. Therefore, family competence, problem solving abilities, acceptance of autonomy and individuality of family members are crucial in the ability of the parents to give a child essential support in CPI. High family competence requires both structure and ability to change it (R. Beavers & Hampson, 2000). By following recommended medical guidelines, parents inadvertently may contribute to their adolescent’s feelings of inadequacy, just by setting limits for self-dependency and self-sufficiency (Vitulano, 2003).

Antecedent factors Mediating factors Consequences

Socio-demographic factors Family functioning mother/youth perspective Mental health HRQoL

Disease severity

Figure 11. Tentative model on psychological well-being of adolescents with chronic illnesses.

Tentative model on psychological well-being in adolescents with CPI is presented in Figure 11. Several models were tested studying the connections between family functioning as a mediator in the relationship between antecedent factors and the psychological outcome in three different CPI groups. The presence and strength of these connections varied considerably between diabetes (Model 1), asthma (Model 2) and epilepsy (Model 3) group models.
First, according to the diabetes path model (Figure 8), older child age, higher disease severity, and changes in the family life (parent’s separation or death) increase the likelihood that an adolescent experience more family functioning problems; the model suggests that the degree of family functioning problems will mediate the relation between the three factors and the decrease of self-esteem/negative self-concept, as well as the negative self-perception of HRQoL in adolescents with diabetes. The relationship between psychological health in children with CPI and disease severity was thoroughly explored earlier (Eiser, 1990). Number of studies showed that youths perceive diabetes as upsetting and hard to manage (Delamater, 2000), taking into the account the difficulties of holding good metabolic control partly due to the decrease of insulin sensitivity associated with puberty (Amiel, Sherwin, Simonson, Lauritano, & Tamborlane, 1986). The latter often lead to the increase in frequency of hypoglycemic episodes and hospitalization. Metabolic control expressed in HbA1c level is the most common indicator of the disease severity in patients with diabetes, it has been shown to predict psychological well-being in adolescents with type 1 diabetes (Hoey et al., 2001). The prospective study of Gustafsson et al. concluded that disturbed family interaction patterns influenced metabolic balance in children during adolescence, but not in younger children (Gustafsson, Cederblad, Ludvigsson, & Lundin, 1987) We did not find such association because the level of metabolic control (HbA1c) was not included as a factor into the model due to the low correlation coefficients with studied parameters.

In diabetes model the most interesting is the appearance of older age as an antecedent factor for changes in family functioning and as a result in HRQoL of adolescents with diabetes. Based on this finding we can speculate that particularly adolescents with diabetes are more vulnerable to challenges and demands of maturation. The older adolescents may perceive diabetes as a restraining and restricting life factor, a threat to future health status and a limitation of their life choices. At the same time parents of the older adolescents tend to provide less disease-specific support and to be less involved in the diabetes care (La Greca & Bearman, 2002) leaving all the disease burden on the adolescents. This is a controversial finding as with maturation adolescents might develop better coping mechanisms and a tendency to more adaptive response to the CPI (Faro, 1999).

Another explanation of the “age” mediating role might be in the disease characteristics, we assume that the chronicity of the illness is perceived differently in different CPI. Diabetes is a lifelong disease, with the continuous treatment requirements, while adolescents with asthma and epilepsy always consider a possibility of improvement or remission. “Child age” as a factor was not included into two other models (Model 3 and 4) due
to the low correlation coefficients with outcome factors in asthma and epilepsy groups.

Second, mothers of adolescents with diabetes perceive single-parent household and changes in a family life as a catalyst for family functioning problems and as a reason behind the increase of behavior-emotional problems in their children. Family structure and the family situation as a key component of the mothers perception of their childrens mental health is an expected finding (Gortmaker et al., 1990).

The third model (Figure 9) examined the associations and pathways underlying relationships of the studied parameters in asthma group. According to the model, being a girl, higher disease severity, and lower SES increase the likelihood that an adolescent experiences more family functioning problems; the model suggested that the degree of family functioning problems perceived by adolescents will mediate the relation between the three factors and mental health outcome resulting in the decrease of HRQoL. Same type of associations was found in Sawyer et al. study (Sawyer et al., 2001). After controlling for the frequency of asthma symptoms a significant relationship was found between children’s reports of the extent to which they are upset and bothered by asthma symptoms (HRQoL), and the functioning of their families. The gender effect on the studied parameters was very small in Model 3. The model suggests that low SES and single-parent household as antecedent factors increase the probability of the family dysfunction reported by asthma group mothers and as an outcome increase the mothers’ reports on behavior-emotional problems of their children. The effect of SES is fairly big in asthma model. Our interpretation of the later finding lies in the ground of family SES itself. It is constructed of three characteristics – parental education, occupation and income level. The first one is crucially important for the ability to explain to the child the importance of the compliance with the health regimens, positively motivating and developing the sense of normality resulting in a better psychological well-being. It has been reported in Apter et al. study of the adult population (Apter, Reisine, Affleck, Barrows, & ZuWallack, 1999) that cultural and socioeconomic factors influence the outcome of the disease, along with severity status. The impact of the income level in our study is disputable taking into consideration that all medical health care (in the hospitals or outpatient clinics), as well as medication expenses are covered by the state for all three studied groups. So in this project the accessibility to health care was controlled to some degree. Same degree of control by surveying patients of specialists was reported by Erickson et al. concluding that the household income was the most consistently associated factor with favorable health outcome for asthma pediatric patients and their caregivers (Erickson et al., 2002).
According to the fourth model (Figure 10), being a boy, lower SES and single-parent household increased the likelihood that an adolescent with epilepsy experience more family functioning problems; the model suggested that the degree of family functioning problems will mediate the relation between the three antecedent factors and self-reported mental health outcome, which causes the negative perception of HRQoL. The strongest effect size was found in family type factor in association with family dysfunction, we explain this finding by the characteristics of our study sample. There were more single-parent families in the epilepsy group compared to other CPI groups.

The absence of “disease severity” factor in epilepsy model is controversial. We can conclude that disease severity is not a decisive factor related to the increase of mental health problems and decrease of HRQoL. Since the outcome in epilepsy group was worse than in the other two CPI groups, there should be other endogenous confounding variables that were not included in this research project. The other explanation might be in the measure of severity used in this project (discussed further in limitations).

In this model the paths between the background factors and mother’s perspective of family functioning were not significant, unexpectedly the family functioning perceived by mothers failed to explain the variation in mental health even reported by the same informant. Therefore, we can conclude that in the studied sample the family environment and functioning from the perspective of mothers with children with epilepsy do not attribute to the differences in adolescent’s mental health. There were no differences in the empirical sources of studied factors between three CPI groups, with the exception of HRQoL.

We tested a tentative model on psychological well-being of adolescents with CPI in three disease groups. By including different areas of psychological well-being into the model we were able to determine whether the model fits equally in explaining HRQoL in three groups of adolescents with CPI. A considerable preponderance of personality characteristics in explaining HRQoL was evident in all three models. The possible explanation might be in the characteristics of the measures used in our study. HRQoL questionnaires, on average, encompass three domains reflecting physical, social, and emotional well-being. We assume that the emotional domain of HRQoL questionnaires might measure similar personality construct as RSES and “Psychological well-being” scale of “I think I am” questionnaire; while the measures of mental health problems (YSR/CBCL) in children do not reflect this personality construct.

The same theoretical model applied across three disease types showed very different fit. The good fit in diabetes group, and a fair fit for asthma group, and even worse for the epilepsy group. We suppose that there are
confounders influencing mental health of adolescents with asthma and epilepsy that we did not include in our tentative model. Maternal mental health and perceived social support might be unexplored variables that could contribute to the variation in adolescent’s mental health mediated by family functioning in our study sample. As well as the intrapersonal factors as cognitive skills, temperament, motivation and problem-solving skills would shed the light on the psychological well-being of adolescents with CPI.

7.1 Limitations

All studies presented in this thesis had a cross-sectional design, and hence do not provide directions for the causal effects. The path analysis, however, allow us to establish which of the potential mediator variables were most important in explaining the overall association with the psychological health of the adolescents as an outcome. The representativeness of the study population might be questioned. The convenience sampling technique used in identifying asthma and epilepsy groups is a major study weakness, while diabetes group are the true population representatives of their age in the city of Arkhangelsk.

The applicability and relevance of the study results can be questioned considering that the data collection was done in 2003. Even though the economic situation in Russia in general is somewhat more stable these days, major economic and socio-demographic parameters and trends remain relatively similar now in 2013-2014 (Federal State Statistics Service 2014). So in my opinion the findings presented here can provide insights into the mental health and HRQoL of children with CPI in regions with developed yet underfinanced health care system and stagnating economy in post-Soviet states.

The selection of healthy controls matching the CPI group only by age is another limitation. There are also other possible covariates that are not studied as: number of other children in the household, the sibship rank, and whether any of these other children had a significant clinical diagnosis, mother mental health, child school performance and cognitive skills. By matching not only basic demographics, but also other important variables, researchers are better able to attribute any differences between the CPI patients and healthy counterparts (McClellan & Cohen, 2007).

Diagnostic subgroups of epilepsy were not defined which might influence results. The severity of the illness was evaluated by an unstandardized measure. It was not possible to find any similar measure of severity applied for all three disorder groups (Stein et al., 1987), therefore we found that the physician’s evaluation of the severity was the most valuable and useful available information.
Although the Achenbach scales have been used in Russia before, there are no norms developed yet, so it was appropriate to use the raw scores instead of norms (T) for the analysis. Note that relationships with covariates such as age may be obscured when norm scores rather than raw scores are used (Grigorenko et al., 2010). The use of multiple informants for the purpose of assessing child and adolescent mental health is viewed as the best practice (Jensen et al., 1999), therefore our study would benefit from also inclusion of fathers or teachers reports.

To reveal the issues of the target population groups, we chose to use disease-specific HRQoL instruments. It was therefore not possible to compare HRQoLs of different disease groups or to conduct a comparison with controls.

There are certain limitation in application and interpretation of a path model. Some potentially relevant variables can be left out or the feedback might be ignored, these specification errors might result in biased estimates. But recognizing the imperfection of the model it is important to remember that it represents a simplification of reality (Acock, 1999).

7.2 Main conclusions

In the scope of the present thesis we assessed and described psychological well-being and family functioning of adolescents with chronic illnesses. Though with much generalization, we believe our findings can be seen as an insight into mental health of children with CPI in post-socialist countries going through transition to market economy and experiencing certain politico-economical instability.

The present thesis was able to demonstrate generally low levels of behavior-emotional problems, depressive symptoms, relatively high self-esteem, positive health-related quality of life, and successful family functioning in adolescent with CPI in Northern Russia. However, we conclude that there is a risk of mental health problems, particularly of internalizing nature, in patients with CPI. These risks were especially pronounced in adolescents with asthma and epilepsy. The only significant risk factor associated with psychological well-being mediated by family functioning in all three CPI groups is single-parent household. Other risk factors contributing to psychological well-being of adolescent with CPI were – child age, child gender, disease severity, family SES, and changes in the family life. High family competence and successful family functioning are protective factors for psychological well-being in CPI adolescents.
7.3 Implications

The findings of this thesis of the psychological well-being of adolescents with diabetes are quite reassuring. Yet, our results should not stop the effort of the healthcare professionals in improving the healthcare system and identifying children and adolescents with diabetes who might develop mental health problems. According to the literature a special attention should be drawn to the eating behaviors and diary habits in this group of adolescents (Helgeson et al., 2007). Also depression has shown to influence long term outcome of type 1 diabetes (de Groot, Anderson, Freedland, Clouse, & Lustman, 2001). While patient adherence, the level of asthma self-management skills, exposure to stress, and depression was shown to have considerable influence on a wide range of asthma outcomes (Rand et al., 2012).

There is evidence to believe that psychological well-being in CPI children from the rural and remote areas might be worse than in their urban counterparts. These differences were found in children with diabetes despite having a similar metabolic control and diabetes knowledge (Cameron et al., 2002). Since the studies reported in this thesis were conducted in the urban area further research is needed addressing this issue.

Although the results of our study showed low levels of externalizing behaviors in children with CPI, we suggest an investigation focused on the health risk behaviors in CPI adolescents is needed. There are evidences suggesting the increase of alcohol consumption in Russian adolescent population (Stickley et al., 2013). Even though these behaviors are considered a normative aspect of adolescent development, risk-taking behavior among CPI youths may cause health effects through substance use, including negative effects on their disease state, potential drug interactions, and improper use of prescribed medications secondary to intoxication (Geist et al., 2003).

There is a paucity of literature focused on evaluation of efficacy of psychosocial interventions for pediatric chronically ill population. There are no reports on interventions in a longitudinal perspective conducted in Russian Federation. However, relying on the international experience we can presume that psychological interventions for disease-related or emotional-behavior problems are effective and that the effectiveness is maintained for at least 12 months following cessation of the interventions (Kibby, Tyc, & Mulhern, 1998). In Kibby et al. meta-analysis it was concluded that children receiving psychological treatment had significantly greater benefits than those who did not. Behavioral and cognitive-behavioral techniques were commonest and most effective type of treatment, suggesting the emphasis on short-term therapies for typical problems experiences by CPI children and adolescents (including disease management, procedural distress and
psychosocial adjustment problems). At the same time interventions based on educational initiatives have demonstrated improved functioning in some pediatric chronic conditions (Gortmaker et al., 1990).

Taking into account our findings we conclude that interventions should be family focused. The mediating effect of family functioning on aspects of psychological health of adolescents differed between the CPI groups but remained relatively strong. Competent, flexible and adaptive family is the goal of any intervention technique, built on partnership and relationships to meet CPI individual members’ needs and strengthen adaptation. In general terms, the family perception of positive support from the health care system (with the focus on the perception of support, which can be different from the intention to be supportive) will have a positive impact on the family’s and the patient’s ability to cope (Geist et al., 2003). The importance of the perceived social support can’t be overestimated and appear as a major factor contributing to general adjustment and psychological health of adolescents with CPI and their families. One of the explanations of elevated mental health problems and lower self-esteem of adolescents with epilepsy might lie in the deficit of social support, and defocused health care system emphasis. One of the important factors with a bidirectional link to psychological health of adolescents with CPI is the compliance with the health regimens. The crucial predictor of the compliance among adolescents with chronic illnesses was support from the health-care specialists (Kyngas & Rissanen, 2001).

A bio-social-psychological framework and interprofessional team work are important conditions for quality improvements in the health care of children with chronic physical diseases.
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