Coping Mechanisms in Short Stature Youth

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Summary

Coping has been considered an important aspect when dealing with health related problems and challenges, such as chronic diseases. However coping mechanisms in children and adolescents with short stature have not yet been examined thoroughly. This thesis examines the effects of gender, age, diagnosis, treatment status and severity of short stature on coping strategies employed by children and adolescents with short stature (Growth Hormone Deficiency and Idiopathic Short Stature). Coping was assessed using the CODI questionnaire and the coping domain of the QOLISSY questionnaire. In total 137 children and adolescents between 8 and 18 years were examined during the retest phase of the QOLISSY study, which took place in five European countries. In total the highest scores of the CODI questionnaire were shown regarding the strategies Acceptance and Wishful Thinking, indicating a frequent use of these strategies in children and adolescents with short stature in this sample. Emotional Reaction as a coping strategy was used the least. Significant gender differences were found on the Avoidance scale. Age differences were found on every scale, with the exception of the Cognitive-Palliative scale. Significant differences were also noted regarding the type of short stature diagnosed and the treatment status of the children and adolescents. Looking at the severity of short stature there were significant differences on the Wishful Thinking domain. Mild significant correlations were found between the CODI and the QOLISSY Coping questionnaire on the scales except Emotional Reaction. A consolidated view indicates that gender, age, diagnosis, treatment status and severity of short stature effects have to be taken into consideration regarding the use of different coping strategies. Especially age differences should be emphasized and a developmental approach to coping might be considered.

1. Introduction

Growth is an important attribute of childhood and adolescents (Schmidt, 2007). It often represents for children a sign of independence and approaching adulthood. In addition to that, height is also an essential indicator for general health and wellbeing (Bettendorf, 2009). This illustrates the importance of growth and height for many children and adolescents as well as their parents. However, approximately 3% of the population is considered to be of short stature (Bettendorf, 2009). Two of the most common and significant types of short stature are idiopathic short stature (ISS) and growth hormone deficiency (GHD) (Cohen, 2008; Hauffa, 2008). Children and adolescents with short stature often have to face a wide range of problems and challenges related to their disease. Previous research indicated that children and adolescents with short stature might potentially be at greater risk for psychosocial problems (Gordon, Crouthamel, Post & Richman, 1982; Holmes, Karlsson & Thompson, 1984; Voss & Mulligan, 1994; Stabler, Clopper, Siegel, Stoppani, Compton & Underwood, 1998; Noeker & Haverkamp, 2000). Still, other researcher suggest that these problems are neither statistical significant nor that the research is conclusive (Ross 2004; Sandberg, 2005; Bullinger, 2009).

This is an important fact to take into account, considering the discussion about the extension of growth hormone treatment beyond children diagnosed with GHD, to include children with ISS as well (Wygold, 2002; Ross et al., 2004; Cohen et al., 2008). Considering these inconclusive results, other concepts might be contemplated to help identify children and adolescents in need for attention. Due to the important influence short stature has on a child's functioning, coping mechanisms may also play a role in how children deal with short stature (Wygold, 2002;,Ross et al., 2004; Visser-van Balen, Sinnema & Geenen, 2006). Although coping has been a research subject for many years, only limited research has been done on the relationship between short stature and coping strategies. This has been aggravated by numerous concepts of coping and psychological adjustment making it hard to find a general basis for research. However, the way in which chronically ill children and adolescents, such as short stature, cope with their short stature might be responsible for the reported variation in health and wellbeing (Petersen, Schmidt, Bullinger & the DISABKIDS group, 2006).

This thesis describes the coping strategies most frequently used in a sample of short stature youth examined by gender, age, diagnosis, treatment status and severity of disease, collected via two coping questionnaires. Chapter one gives a short introduction into the topic and leads over into chapter two which presents the theoretical background of this thesis. Chapter 2.1 deals with the basics of short stature and part 2.2 goes into the theory of coping. Following this, chapter three illustrates the methodology, first presenting the objective of this thesis and giving an overview over the QOLISSY study and then describing the instruments used and the analysis procedure. Chapter four presents the results oriented at the hypotheses and these results are then discussed in chapter five. The thesis is then completed with a conclusion and an outlook.

2. Theoretical Background

This chapter is composed of two main aspects of the theoretical background for this thesis. The first part deals with the main aspects of short stature and following that, the concept of coping and the possible implications for children with a chronic disease are presented.

2.1 Short Stature

The following part is structured into six subdivisions. First a definition and epidemiology for short stature is presented. In addition to that possible causes for short stature are shown. In this context two types of short stature, idiopathic short stature and growth hormone deficiency are elaborated on in more detail. Furthermore possible treatment options are presented and psychosocial consequences of short stature are discussed.

2.1.1 Definition/Epidemiology

Height can be influenced by many factors, genetic, metabolic, psychological and environmental (Li & Power, 2004; Hauffa, 2008). Normal height in a population is best defined and expressed as a standard deviation score (SDS), within ± 2 standard deviations from the mean height for age and gender. Short stature is by this definition a body height of an individual more than 2 SD below the norm of a population in reference to age and gender and less than 1.5 SD of the mid-parental height (Cohen et al., 2008). By using SDS instead of height in cm it allows for comparisons between countries, with different average heights, between different age groups and sexes (Lindsay, Feldkamp, Harris, Roberson & Rallison, 1994). The German Association for People of Short Stature and their Families (BKMF, 2011) estimates that 100.000 people in Germany live with short stature.

In a similar definition short stature is defined as a height below the 3rd percentile in reference to country, age and gender (Bettendorf, 2009). By this definition 3% of the population of any country is considered to be of short stature. Consequently 125.000 children are born each year in the EU with some type of short stature (QOLISSY manual, 2011).

2.1.2 Aetiology

Forms of short stature are distinguished between primary and secondary types of short stature. Primary short stature is the result of a prenatal dysfunction or an intrinsic (genetic) abnormality in the skeletal system, whereas secondary short stature is a result of a defect in the factors required for growth (Hauffa, 2008). Table 1 shows the variety of different causes that may result in short stature.

PRIMARY SHORT STATURE	SECONDARY SHORT STATURE
Genetic or familiar short stature	Constitutional delay of growth
Intra uterine growth retardation	Malnutrition
Chromosom abnormalities (e.g. Ulrich-Turner Syndrom, Trisomie 21)	Gastrointestinal abnormalities
Developmental anomalies (e.g. Russel-Silver-Syndrom, Noonan-Syndrom)	Psychosocial deprivation
Metabolic defects	Renal insufficiency
Skeletal dysplasia	Cardiovascular diseases
	Lung diseases
	Anemia
	Endocrine deficiencies (e.g. GHD)
	Medication
	CNS radiation

Table 1: Causes of short stature (see Hauffa, 2008)

In addition to the causes listed in the table above short stature can also be classified as idiopathic, when no underlying pathology can be attributed to the short stature (Cohen et al., 2008). Due to the lack of knowledge regarding the cause of idiopathic short stature it cannot be assigned explicitly to primary or secondary short stature.

This thesis focuses in particular on idiopathic short stature (ISS) and growth hormone deficiency, as children with GHD and ISS usually do not have other impairments of health and wellbeing (the QOLISSY manual, 2011).

2.1.3 Growth Hormone Deficiency

Growth hormone deficiency is the most common endocrinological causes of short stature. Children and adolescents with GHD have a complete or partial absence of

growth hormones or by secretion of abnormal GH or lower levels of growth factors such as the insulin like growth factor (IGF-I), on which the GH depends (Richmond & Rogol, 2008). This is often caused by a malfunction of the growth hormone producing pituitary gland (Binder, 2002).

GHD can be congenital, acquired or idiopathic. A congenital GHD is seen in one of 3500 birth (Hauffa, 2008). It can be caused for example by genetic defects or deformities of the pituitary gland. Reasons for an acquired GHD can be a traumatic birth event, tumours, radiation of the central nervous system, a traumatic brain injury or infections (Hauffa, 2008). Idiopathic GHD is the most common form where the cause of insufficient secretion of GH or the lack thereof is unknown.

According to Hauffa (2008) characteristics of children with GHD are:

- proportionate short stature with delayed bone aging
- a small facial portion of the scull compared to the neurocranium with a small chin and a shrunken root of the nose
- small hands and feet
- weakly developed muscles
- relative obesity
- thin skin and hair
- high voice
- a small penis
- delayed puberty

2.1.4 Idiopathic Short Stature

Idiopathic short stature is a diagnosis of exclusion (Lee, 2006). It is defined as a condition in which the affected person has a height below -2 SD of the average height in reference to age, sex and population group. But contrary to other causes of short stature no apparent medical cause can be found (Ranke, 1996; Cohen, 2008). The affected children present a normal size at birth, normal or low growth velocity and no evidence of an underlying pathology (Ranke, 1996). ISS represents a heterogeneous diagnosis group in which no causes could be found with current diagnostic tools (Schmidt, 2007). This categorisation of ISS also includes children and adolescents with familiar short

stature and constitutional delay of growth (Ranke, 2001; Cohen, 2008). Treatment options for children and adolescents with ISS are limited in the European Union (Savage, 2009). According to Schmidt (2007) without a cause of short stature there is no basis for an adequate treatment. Growth hormone therapy for patients with ISS has not been approved by the European Medicines Agency (Savage, 2009). However psychological and medical interventions are currently discussed in Europe, but further studies are needed on this topic (Visser-van Balen, Geenen, Kamp, Huisman, Wit & Sinnema, 2007).

2.1.5 Treatment

Short stature can be diagnosed early in childhood and therefore should be treated early (Bettendorf, 2008). The children are often noticed in routine pediatric examinations to be abnormally short. This can be determined by reference to country specific growth curves for example the WHO growth curves. Even though short stature is often diagnosed early there are only limited treatment options available. In 1960 the human pituitary growth hormone became available for treatment of GHD which was first taken from corpses and therefore limited in its application. In 1985 the biosynthetic human growth hormone (rhGH) became available (Sandberg, 2005). This opened the possibility to treat a high number of patients each year and increase the adult height of children with GHD.

Treatment with rhGH is mostly started right after the diagnosis is made. The effected children have to be injected every day with the growth hormone via a pen until the end of their linear growth. This is reached at a bone age of 16 years in boys and 14 years in girls (Bettendorf, 2009).

In addition to a simple optimisation of height an increased quality of life is considered to be an essential goal of GH treatment (Cohen et al. 2008; Chaplin, 2011), due to the fact that short stature is suggested to have a negative influence on the child's development and psychosocial functioning (Stabler et al., 1994; Voss, 1994; Noeker & Haverkamp, 2000).

This argument is also part of the discussion regarding the treatment of children with idiopathic short stature with growth hormones. Even though the hormone production reaches a normal level, the growth receptors cannot absorb GH the same way normal receptors do (Hauffa, 2008). Researches in the past tried to increase the

height of children with idiopathic short stature by treating them with growth hormones as well. However studies show that an increased adult height can be reached even thought the adults with ISS will still be shorter than their peers (Finkelstein, Imperiale, Speroff, Marrero, Radcliffe & Cuttler, 2002; Bryant, Baxter, Cave & Milne, 2007; Bullinger et al., 2009). Since the benefits, children and adolescents receive due to GH treatment, are only minor in regard to height gain, other potential benefits have to be examined in order to conduct weather idiopathic short stature should be condoned (Bryant et al., 2007). However the European Medicine agency (EMA) has not approved a treatment with GH for children and adolescents at the moment.

This suggests that without a cause of short stature there is no basis for an adequate treatment and no medical treatments are available for children and adolescents with ISS (Schmidt, 2007). Therefore psychosocial interventions have been suggested not only for children with ISS, but also GHD in order to strengthen the effected children and their resources, including the family (Noeker, 2009). In psychosocial interventions coping is often an important topic and strategies are used to strengthen the self concept and self assertiveness. Psychosocial interventions also help to investigate the potential psychosocial problems and challenges and could help to determine whether psychological interventions can increase the quality of life in children and adolescents with short stature (Noeker, 2009).

2.1.6 Psychosocial aspects of Short Stature

Short stature is not only connected to physical problems, but is also suggested to be a risk factor for psychosocial problems (Stabler et al., 1994; Voss, 1994; Noeker et al., 2000; Magnusson, Gunnell, Tynelius, Smith & Rasmussen, 2005; Abe et al., 2009). The psychological impairments have been widely investigated in research; however the evidence is inconclusive weather short stature has a significant impact on wellbeing and psychosocial functioning (Sandberg, 2005; Bullinger et al., 2009). Studies have shown the impact of disparity in height on the social perception of their environment including their peers (Sandberg & Voss, 2002). Psychosocial effects of short stature have been reported to include negative comparisons with peers, social stigmatization due to height-related stereotypes (Sandberg & Voss 2002, Bullinger et al., 2009) and social exclusion. These aspects can have a substantial influence on the self-esteem and behavior and

might account for the child being a target for bullying at school or in other social settings (Voss & Mulligan, 2000; Harter, 2001). Short statured people were also recorded to present with higher rates of social anxiety, aggressiveness, immaturity and depression (Sandberg & Colsman, 2005; Abe et al., 2009, Bisacchi et al., 2011). Wheeler et al. (2004) reported short statured children and adolescents as having lower scores on intelligent tests and academic achievement, but within the normal scale. However efforts to measure psychosocial aspects of short stature have been proven to be difficult (Sandberg, Bukowski, Fung & Noll, 2004; The QOLISSY manual, 2011). Studies also report that some children of short stature experience chronic psychosocial stress, even though these experiences usually do not result in clinical significant problems of psychosocial adaptation (Sandberg, 2005).

In contrast many studies contradict these results, reporting levels of behavior problems in the normal range (Kranzler, Rosenbloom, Proctor, Diamond & Watson, 2000; Voss & Sandberg, 2004; Sandberg & Colsman, 2005b; Visser-van Balen et al., 2006, Lee et al., 2009). These in part contradictory findings could be uncovered in the review of the literature and suggest the need for further research as influence of short stature on psychosocial competencies remains unclear, providing an occasion to investigate why some children and adolescents experience chronic psychosocial stress and others deal with their condition without problems.

Therefore it is important to explore how children and adolescents cope with their short stature and which coping strategies they use in order to assess which factors might help them to develop normally.

2.2 Coping

The following part gives a definition of coping and a well-established coping model is presented. Furthermore a developmental approach to coping is discussed.

2.2.1 Definition

Coping has been an important topic in research for decades. The idea goes back to the psychological concept of "defense mechanisms" of Anna Freud (1936), who characterized them as efforts to tackle unpleasant thoughts and feelings, and realized that there are individual differences of how people deal with these.

Since then the definition of coping has changed a lot over the years, but one of the most influential people in the field of coping research Richard Lazarus, together with his colleague Susan Folkman (1984) defined Coping as "constantly changing cognitive and behavioral efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person" (Stress, appraisal and coping, 1984, p.141). Another important characteristic of coping is, that it includes all generated efforts to deal with a stressful situation, independent of their value and effectiveness (Lazarus & Folkmann, 1984). This means coping can take place even though the desired result is not achieved and can even have a negative impact on the situation.

2.2.2 Coping with a chronic disease model

Coping is used not only in stressful daily situations, but is especially needed when dealing with a disease (Warschburger, 2009). In such situations the person is often exposed to many stressful internal and external demands and has to find ways to deal with them. Many factors influence this coping process. It is not only dependent on the medical characteristics of the disease, but also on psychological factors for example personality traits (Zbinden & Perrez, 2002).

One of the well-established models to describe coping mechanisms is the transactional model of stress and coping by Richard Lazarus (1978). This model shows stress situations as complex interaction processes between the demands of the situation and the affected person. In contrast to other earlier models he put the focus not on the nature of the stressor, but on the subjective appraisal by the affected person (Lazarus & Launier, 1978). This emphasizes the individual differences in relevance attributed to the stressor. The transactional nature of the model is shown by the appraisal process between the stressor and the coping outcome.

Meas, Leventhal and de Ridder (1996) presented an extended model for coping with a chronic disease (figure 1) based on the transactional model of Lazarus. According to this model the coping process is influenced by many factors. Those may be life events in general not necessarily related to the disease itself (e.g. divorce of parents). Other important events however can be disease related. In addition to that disease and treatment characteristics can influence the appraisal and thus the coping with a disease

(Meas, Leventhal & de Ridder, 1996). An important aspect in this category is for example the controllability and changeability of a disease (Zbinden & Perrez, 2002). Coping mechanisms are also significantly influenced by demographic characteristics such as sex, socioeconomic status and age (Zbinden & Perrez, 2002).
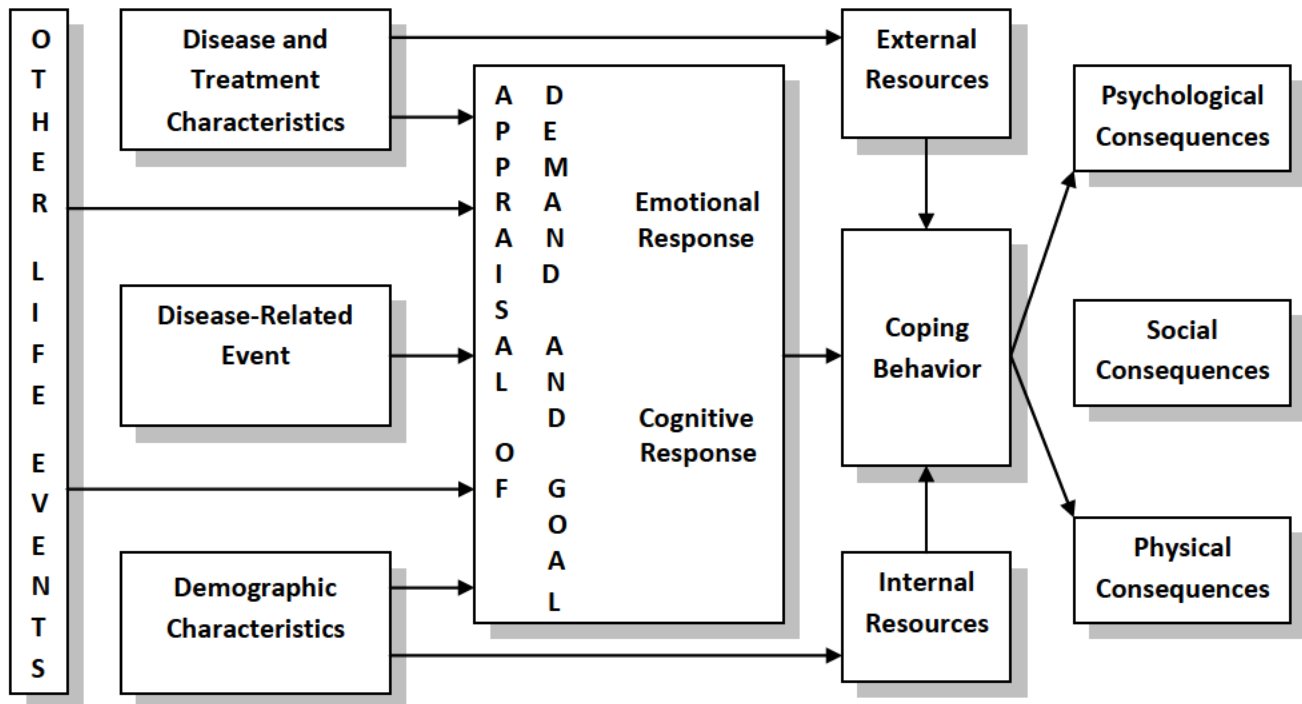

Figure 1: Coping with a chronic disease model (see Meas et al., 1996)

These influences are then appraised regarding the demands and goals. Lazarus (1966) divides the appraisal of the stressor in primary appraisal and secondary appraisal. In the primary appraisal the situation is evaluated as positive, irrelevant or potentially harmful. If a situation is regarded to be irrelevant no adaption processes are activated. In the secondary appraisal the resources to solve the problem are being assessed. If these resources are measured to be not sufficient a coping strategy is considered. Meas et al. (1996) divide these potential responses in emotional and cognitive response. Lazarus however sees an additional type of response: the problem-oriented response (Lazarus, 1999). The problem-oriented response is characterized by strategies to actively influence the problematic situation or refrain from influencing it or adapt to the circumstances. Emotional responses serve to relieve emotional stress induced by the stressor. Cognitive responses also called "reappraisal" are used to

reevaluate the situation and the relationship between the environment and the person and therefore enable a new interpretation of the stressor (Lazarus, 1999).

In addition to that external and internal resources have an influence on the resulting coping behavior. External resources include social support and work environment, whereas internal resources are person oriented including resilience and locus of control. Combined these factors result in situation-dependent coping behavior (Meas et al., 1996). This coping behavior can be categorized in different types of coping strategies, which are situation-dependent, but have also been shown to tend to be used with different frequency in affected people dependent on personality (Zimmer-Gembeck & Skinner, 2011).

Cohen and Lazarus (1983) distinguish between three kinds of consequences resulting from this process: psychological, social and physical consequences.

2.2.3 Developmental approach to coping

Due to the increasing interest in coping over the years countless numbers of publications are available and a number of coping questionnaires have been developed (Aldwin & Revenson, 1987). The primary focus of most of this research however has dealt with coping mechanisms in adults. Research of coping in children began to take off in the 1980's. Since then many coping strategies in children and adolescents have been identified, underlining the importance of this concept in many parts of children's life's (Aldwin, 2007). However fewer efforts have been made to integrate a developmental approach to the coping of children and adolescents, presumably due to the diversity of developmental issues to be considered (Schmidt, Petersen & Bullinger, 2003).

Children and adolescents have to master a variety of developmental tasks while growing up (Havighurst, 1972). These are grouped into certain phases of development: early childhood, preschool age, middle childhood and adolescents (Papastefanou, 2009). This thesis discusses children and adolescent from the age of 8 to 18, so the relevant phases are middle childhood and adolescents.

Middle childhood is characterized through several developmental tasks the child has to fulfill: gain social competencies, learn to read, write and calculate, the development of moral values, to learn appropriate role behavior and the gain some kind of independence (Havighurst, 1972). They also learn to regulate their emotions through

cognitive strategies and learn to deflect uncontrollable stressors (Papastefanou, 2009). The beginning of school is one of the main challenges children have to face.

Adolescents confront the children with different developmental tasks: forming of relationships with peers of both sexes, detachment from parents, preparation for work life, achievement of socially responsible behavior and development of identity (Havighurst 1972; Erikson, 1972). These processes go along with a number of emotional problems, often resulting in self-doubt. But they also improve their mental capacities and gain new ways of reflecting and problem-solving. Emotional regulation processes reinforce themselves and children learn that distraction is a way to momentarily relief stress (Papastefanou, 2009).

All children have to go through these developmental tasks and use various coping strategies doing so. However children with chronic diseases face additional challenges in their daily life's. They have to simultaneously manage to reach their developmental goals, which they share with healthy children, and to cope with the problems caused by their disease (Reich, 2010). Children in middle childhood develop a new understanding of disease due to their growing cognitive abilities. They have to learn to deal with disease related aspects of their life more responsible than before. Due to their illness and the often involved intensive treatment limitations in their daily life problems arise under which the children suffer. Therefore they are more likely to have problems with concentration, often resulting in a decreasing performance at school. In addition to that they might be confronted with social exclusion in school due to their illness, which can result in self-doubt and uncertainty (Papastefanous, 2009).

Dealing with a chronic disease during adolescents can aggravate problems characteristic for adolescents. They feel limited by their illness-related responsibilities (e.g. regular injections). In addition to that engaging in risky behavior common for adolescents (e.g. alcohol consumption) can pose increased health risks, and therefore social exclusion may result. Problems can especially occur if the illness is externally recognizable (e.g. short stature). The disease often becomes one of the defining characteristics of the affected person. The sense of self-worth can be in danger and in extreme cases a rejection of the own body can occur (Papastefanous, 2009).

Due to the significant effects chronic conditions (e.g. short stature) have on functioning of children and adolescents, coping styles may play a role in how the

children adapt to the condition (Marsac, Funk & Nelson, 2006). A recent review (Zimmer-Gembeck & Skinner, 2011) examined coping strategies used by children and adolescents from a developmental standpoint. They identified four most commonly used families of coping with each group representing a functioning set of ways of coping. These were then examined according to the four developmental phases of childhood and adolescents.

The review revealed that children in middle childhood start to use cognitive strategies to cope with upcoming problems. They are also more capable to use adaptive coping strategies. In addition to that support-seeking becomes more variable as they seek support from different sources and escape becomes less apparent (Zimmer-Gembeck & Skinner, 2011).

In adolescents the ways of coping become more sophisticated. As they grow older they are more capable of reflecting on a situation and combine strategies to cope with problems. Also distraction tactics become more diverse as they use behavioral and cognitive strategies to escape, often from situations or problems uncontrollable to them. In addition to that they are more capable of emotional self-regulation (Papastefanous, 2009; Zimmer-Gembeck & Skinner, 2011).

Besides the developmentally related age differences in coping styles, other factors also contribute to differences in coping styles. Gender also influences the choice of coping strategies. In general, woman use emotional and avoidance strategies more often, whereas men use distancing and cognitive strategies (Folkman & Lazarus 1980; Tamres, Janicki & Helgeson, 2002; Matud, 2004). This might be an important factor in coping differences between sexes, because coping using Avoidance and Emotional Reaction strategies is often associated with greater distress, whereas problem focused coping is reported to cause less distress (Maes et al., 1996).

Even though research on children and adolescents with short stature has been published at length, research on coping mechanisms and short stature has been very limited. Most articles deal with psychosocial risk factors, but remark little on coping strategies to deal with these problems. The few articles dealing with coping and short stature often only examine how children and adolescents cope with the growth hormone treatment and not the short stature in itself (Leibermann, Pilpel, Carel, Levi & Zadik, 1993; Ross et al., 2004).

3. Methodology

The following segment deals with the objective of this thesis and will then give an overview of the QOLISSY project, including sample recruitment. After that the instruments for data collection will be introduced and the analysis procedure will be illustrated.

3.1 Objective of the thesis

Deriving from the theoretical background a few questions arise regarding the coping mechanisms in children and adolescents with GHD/ISS. One of the main objectives is to determine what coping strategies are used by children and adolescents with GHD/ISS in this sample. In addition to that the objective of this thesis is to examine if there are significant differences in the coping strategies of different sub-groups. The following hypotheses will be investigated:

- Coping strategies of children and adolescents with GHD and ISS differ regarding the sex.
- Coping strategies of children and adolescents with GHD and ISS differ regarding age.
- Coping strategies of children and adolescents with GHD and ISS differ regarding the diagnosis.
- Coping strategies of children and adolescents with GHD and ISS differ regarding the treatment status.
- Coping strategies of children and adolescents with GHD and ISS differ regarding the parent-child relationship.
- Coping strategies of children and adolescents with GHD and ISS differ regarding the severity of short stature.

The examination of these hypotheses will be done using two different questionnaires. In addition to that a correlation of aspects of the two questionnaires will be done.

3.2 Quality of Life in Short Stature Youth –

The QOLISSY Project

In the following part the European research project "**Q**uality **o**f **L**ife **i**n **S**hort **S**tature **Y**outh – the QOLISSY Study", in which data collection for this thesis took place, will be presented.

3.2.1 Overview over the project

The European QOLISSY study started in 2009. The aim of the study was to develop a disease specific, health-related quality of life (hrQoL) instrument for short statured children and adolescents. The questionnaire specifically targets children and adolescents with idiopathic short stature and growth hormone deficiency and was developed to measure the impact of the short stature on the children's wellbeing and functioning (the QOLISSY Study protocol, 2009).

The QOLISSY questionnaire should be applicable to children between the ages of 8 to 12 and adolescents from 13 to 18 years. In addition to the self-reported questionnaire a proxy version was developed for the parents of affected children and adolescents from the age of 4 to 7, 8 to 12 and 13 to 18 years (the QOLISSY manual, 2011).

The QOLISSY study formed out of the collaboration of internationally recognized quality of life researchers and pediatrician and drew from the expertise from two previous hrQoL studies involving children and adolescents, namely the KIDSCREEN (The KIDSCREEN group, 2006) and DISABKIDS (The DISABKIDS group, 2006) studies.

Five European countries took part in the QOLISSY project. The international project coordination fell to Ms. Prof. Monika Bullinger from the University Medical Centre in Hamburg-Eppendorf. The four other participating countries Sweden, France, Great Britain and Spain each had a national project coordinator responsible for the organisation within the country. Recruitment of the participants was done via clinical centres in the countries (the QOLISSY Study protocol, 2009).

COUNTRIES	NATIONAL CENTER
Germany	University Medical Centre Hamburg-Eppendorf Prof. Dr. Monika Bullinger & Dr. Julia Quitmann
Sweden	The Queen Silivia Children's Hospital Dr. John Eric Chaplin
France	Hôspital des Enfantes Dr. Emmanuelle Mimoun
Great Britain	The University of Edinburgh Prof. Mick Power, PhD
Spain	IMIM/Insight Consulting and Research Michael Herdman

Table 2: Participating countries and national centres

The QOLISSY questionnaire was developed using a cross-cultural approach, being simultaneously developed in five languages. The development procedure followed generally excepted guidelines for questionnaire development of the Federal Drug Administration (FDA) and the "Guidelines for PRO instrument development" and followed the recommended steps in questionnaire development, which will be presented in the following paragraphs (Patrick, Burke, Powers, Scott, Rock & Dawisha, 2007).

Literature Research

First a review of existing literature was done to investigate potential problem areas for children and adolescents with short stature and give an overview over existing instruments. The literature review revealed an insufficiency of disease specific instruments for children and adolescents and especially for short statured youth (Brütt et. al, 2009).

Focus groups

In order to create a questionnaire as close as possible to the problems and challenges faced by short stature children and adolescents a close integration of people affected by the disease is necessary (Bullinger, Schmidt, Petersen, Erhart & Ravens-Sieberer, 2007). They serve as experts for their disease and can be a helpful addition of knowledge in addition to existing literature.

For this purpose group discussions are considered to be more effective than individual interviews (Bullinger et al., 2007). They are used to identify relevant concepts

of quality of life for short statured children and adolescents and generate items and statements used as the base for the questionnaire development.

A focus group manual, with prepared questions for the semi-structures group interviews, was created to ensure comparable procedure of the focus groups. In total 200 participants, i.e. 86 children and 114 parents were recruited via the clinical centres in the countries. After consenting to participation in the study they were invited to join the focus groups. These group discussions were audio recorded for future transcription. In addition, several other questionnaires regarding the health and wellbeing of the children were distributed to gain further knowledge from the participants.

Item generation

The audio recordings then were transcribed and a qualitative analysis of the transcriptions took place. Statements relevant to the topic were identified, translated into English and written on cards. These cards were then on an international meeting of all participating countries again analyzed and relevant categories were formed and a preliminary item list was generated. The categories were the foundation for the development of a conceptual model containing three main categories of quality of life (QoL): physical QoL, social QoL and emotional QoL. In addition to that moderating aspects have been identified: Coping, Attitude, Treatment and Effects on parents. A more detailed description of the coping segment which will be examined in this thesis can be seen in part 3.3.2.

In addition, the items were written down in English, followed by a translation into the project languages (German /French /Spanish /English /Swedish) and back. This process of back- and forth-translation is seen as the best method to develop a questionnaire parallel in different countries (Bullinger, Power, Aaronson, Cella & Anderson, 1996).

Pilottest / Cognitive Debriefing

The next step was to perform a pilot study in order to deduce which items should be modified or excluded from the questionnaire. In total 205 people participated in the pilot test. The pilot questionnaire was given to children and parents to be filled out and afterwards participants were interviewed individually regarding relevance, clarity,

sensitivity and importance. These results were analyzed regarding psychometric properties. In a discussion on international level the questionnaire was modified.

Field test and retest

The next phase in the questionnaire development aimed at testing the psychometric properties of the modified questionnaire in order to prepare for final use of the QOLISSY questionnaire. Again after informed consent was obtained 268 children and adolescents and 317 parents received the questionnaire using the approach considered to be most appropriate for each country. Families who consented in continued participation then received the questionnaire again two weeks later in a retest. In total 137 children took part in the retest. A more detailed description of the retest procedures can be found in section 3.2.2.

Application in further research

The developed cross cultural QOLISSY questionnaire is going to be translated into further languages. Validation studies with the Netherlands, Belgium, Italy and the United States of America are already being conducted. In addition to that the questionnaire could be implemented in clinical research or modified to specifically fit other types of short stature.

3.2.2 Sample recruitment

The data which is the basis of this thesis was attained during the retest phase of the QOLISSY study. In addition to the development of the QOLISSY questionnaire, the retest phase was also used to acquire additional data on children and adolescents with GHD and ISS. For this purpose in addition to the QOLISSY questionnaire a battery of other questionnaires was also distributed in phases of the study. One of these questionnaires was the **Co**ping with a **Di**sease (CODI) questionnaire which is, besides the QOLISSY questionnaire, the main instruments for this thesis (Petersen et al., 2004).

As mentioned above the main objective of the retest phase was to test the QOLISSY's psychometric properties and test-retest reliability. The testing was performed in all five countries. Potential participants were identified by clinicians in the clinical centers and informed about the aims and the procedures of the study. They were

then asked whether or not they are willing to participate in the study. If interested children as well as the parents were asked to sign an informed consent form. The families were than given the QOLISSY questionnaire and additional questionnaires such as the CODI. When returning the field test questionnaire families were asked to fill out the same questionnaire again for the purpose of the retest. This was accepted by more than 50% of the participants, who returned the second questionnaire within 10 days.

3.2.2.1 Inclusion and exclusion criteria

In the following paragraph the criteria for inclusion and exclusion of participants into or from the QOLISSY study are being presented. They are following the international manual of the QOLISSY study (2011).

Children and adolescents with a diagnosis of growth hormone deficiency (GHD) or idiopathic short stature (ISS), and consequently a height of below -2SD at the beginning of treatment, between the ages of 8 and 18 were included in the study. The perspective of younger children from 4 to 7 years old were only included through proxy questioning of the parents, due to developmental considerations. The perspective of the parents however will not be considered in this thesis. In addition to that, the participating children had to be able to answer the questionnaires, regarding their cognitive abilities. A further inclusion criterion is a sufficient knowledge of the respective language of the country in which the children and adolescents participate in the study. Furthermore they had to consent to participating in the study voluntarily and a signed consent form of the children and adolescents as well as of one parent or legal guardian has to be filled out.

Children and adolescent with other forms of short stature have been excluded from the study as well as children with other chronic diseases, which could influence their quality of life. Children who did not consent to participate were excluded from the study as well.

3.3 Instruments

In the following paragraphs the two questionnaires central to the topic of this thesis will be presented.

3.3.1 CODI questionnaire

The CODI questionnaire was developed for the DISABKIDS project (Petersen et al., 2004), which aims at improving the quality of life of children with a chronic condition (the DISABKIDS group, 2006). In the following segments a description of the CODI questionnaire is given and the scoring is described.

3.3.1.1 Instrument description

The CODI questionnaire was developed in a similar approach as the QOLISSY questionnaire (see 3.2.1). First focus groups were done, in order to generate a preliminary item list for pilot testing. After improvements to the instrument were integrated the psychometric properties were assessed in field- and re-testing. The CODI questionnaire was developed simultaneously in six European countries (Austria, Germany, Greece, Sweden, the Netherlands and Great Britain), including children with seven different chronic diseases (Arthritis, asthma, atopic dermatitis, cystic fibrosis, cerebral plasy, diabetes mellitus and epilepsy) (Petersen et al., 2004). After refining of the items 28 items on a five-point Likert scale were left for the final CODI questionnaire. These 28 items represented six coping strategies, listed here with one example per strategy: Avoidance: "I try to ignore my illness", Cognitive-palliative: "I realize that thinks could be worse"; Emotional Reaction: "I am frustrated"; Wishful Thinking: " I hope my illness will go away"; Acceptance: "I got used to my illness"; Distance: "I think my illness is no big deal. An additional question ascertains how the children cope with their illness in general. Receiving the questionnaire children got an instruction on how to answer the questionnaire: "Think about situations when you were anxious or stressed because of your illness. Below you will find a list of some common reactions children and teenager have as they cope with illness. Please tell us how typical these actions or thoughts are in relation to your illness" (see Appendix I). On the answering scale the children and adolescents could rang their answers from "never" to "almost never", "sometimes", "Almost always" and to "Always". These aspects indicate how frequent they apply a certain coping strategy.

3.3.1.2 Scoring of the questionnaire

Mean scale scores are calculated as the sum of the answers given by the participants (ranging from 1 to 5 per item) divided by the number of items of each scale. If more than

one item of the scale is missing, the scale score is not computed. The CODI scale scores are then transformed from raw scores to a scale from 0 to 100 scale. A higher score is associated with a more frequent use of the coping strategies

With regard to the internal consistency, Cronbach's alpha ranges from α= .72 to .88 for the different coping scales (Petersen et al., 2004).

3.3.2 QOLISSY questionnaire

The development of the QOLISSY questionnaire was the main goal of the "Quality of Life in Short Statured Youth" study. In the following segments a description of the QOLISSY questionnaire is given and the scoring is described.

3.3.2.1 Instrument description

The QOLISSY questionnaire was developed according to the steps presented in section 3.2.1. The QOLISSY questionnaire was developed as a patient reported outcome instrument for short stature children and adolescents with GHD or ISS. The age range includes children between the ages of 8 to 12 and adolescents 13 to 18 years old. In addition to that a parent's proxy version was developed for the age groups 4-7, 8-12 and 13-18 (the QOLISSY manual, 2011).

The QOLISSY questionnaire consists of three core dimensions (Physical QoL, social QoL and emotional QoL) with 22 Likert–scaled items, and three additional domains (Coping, Attitude and Treatment) with 28 Likert–scaled items. Results of the field test showed satisfactory internal consistencies of these scales, ranging from $\alpha = .81$ (Physical) to $\alpha = .87$ (Social) (the QOLISSY Study group, 2011).

On the answering scale the children and adolescents could express their degree of consent/approval from a 5 point scale ranging from "not at all/never", "Slightly/seldom", "Moderately/quite often" to "very/very often" and "Extremely/always" (see Appendix II). The three core dimensions can be combined, to create a general score for HrQoL for children and adolescents with short stature.

The Coping segment of the QOLISSY questionnaire consists of 10 items representing five general ideas of Coping, which were identified in accordance to the focus groups. These strategies are Acceptance, Ignoring/locking out, Aggression/acting out, Denial, and seeking/receiving support. However, the 10 items are not specifically

organised to represent these ideas. Item examples are: "I tell myself it is Ok to be short", "When I feel bad about my height I talk about it to family and or friends" and "When I feel bad about my height I try to forget about it" (see Appendix II).

3.3.2.2 Scoring of the questionnaire

The interpretation of the QOLISSY scores is based on the statistical distribution of scores with regard to specific reference groups (the QOLISSY Study group, 2011). The scores are transformed from raw scores on the five-point Likert scales to scores from 0 to 100 and examined on the individual dimensions (physical, social, emotional) as well as a QOLISSY quality of life total score, including all three domains. Higher scores indicate a higher quality of life. These scores can be compared to reference scores in the QOLISSY manual, respecting country, age and gender differences. A total score can be calculated for each domain as well. A high score on the Coping domain indicates a more frequent use of coping strategies (the QOLISSY manual, 2011).

3.4 Analysis procedure

Data was analysed using the Statistical Package for Social Scientists (SPSS), version 18.0. In order to determine if there are significant differences in the use of coping strategies between the examined groups a comparison of the means of the groups has to be carried out. This can be done using parametric or nonparametric tests. For parametric testing a number of requirements have to be fulfilled: The data has to have a normal distribution, the samples have to be roughly of equal size, homogeneity of variance has to be present and the participants have to be independent from each other (Field, 2009). If those requirements are not met nonparametric tests are more accurate (Field, 2009).

Looking at the data the requirements for parametric testing are in general met. Sample sizes are overall big enough (N ≈ 130) so that normal distribution can be assumed. In samples of this size the distribution of the data only has an extremely slight impact on the tests (Tabachnick & Fidell, 2007; Burdenski, 2000). Also the groups are of roughly the same size and looking at variance homogeneity data reached scores above 0.05 on the Levenes' test of homogeneity, indicating that the data could be analysed parametrically. Therefore t-tests were used to test weather coping strategies differ between the different examined aspects e.g. gender, age, diagnosis and treatment

status. An univariate analysis of variance (ANOVA) and post hoc test was used to examine differences in the coping strategies of the three groups of severity of short stature (mild, moderate and severe). In addition to that effect sizes were calculated using Cohen's d. The effects were interpreted with a d of around 0.1 as a small effect; an effect of around 0.5 is assumed to be a medium effect and an effect of 0.8 to infinity being a large effect (Cohen, 1988; Field, 2007). Negative effect sizes were reported positive, to facilitate readability. Negative numbers only indicate directions of the differences between the groups, which in this case is done by showing the means, and do not impact the size of the effect (Field, 2007).

In addition to that a control of the results was done, in which the same analysis was performed, but with the assumption of different conditions of the data. This was done to verify the results of the parametric testing, assuming the above mentioned requirements of parametric testing were not fulfilled and therefore nonparametric testing was performed. The differences in coping were tested using Mann-Whitney-U and Kruskal-Wallis tests. Effect sizes were calculated using Pearson's r. The effects were interpreted with an $r = 0.1 - 0.23$ as a small effect size, $r = 0.24 - 0.36$ a medium effect and an r of 0.37 or higher as a large effect size (Cohen, 1992).

Statistical significance was defined at a level of $p \leq 0.05$. The relationship between CODI coping strategies and QOLISSY Coping was investigated with the Pearsons's Correlation between scales.

4. Results

In chapter four a description of the sample will take place and the results of the statistical analysis will be presented.

4.1 Sample characteristics

The sample was composed of 137 children and adolescents from the age of 8 to 18 years. In average the participating children and adolescents were 13.18 years old (table 3). The children and adolescents were sorted into two groups regarding their age. 43.8% of the participants were between 8-12 years and adolescents between 13 and 18 years represented 56.2 % of the sample. One of the study participants aged 7 was considered an exception, due to him already visiting school and his ability to independently read and answer the questionnaire despite his young age. He was therefore included in the age group 8-12 years.

Male participants were slightly overrepresented with 56.2 % of the total participants (table 3). 43.8% of the children and adolescents were diagnosed with a Growth Hormone Deficiency (GHD) and 56.2% of the participants with Idiopathic Short Stature. 60.6% of the children and adolescents were being treated with growth hormone or had been treated in the past. The remaining 39.4% of the participants did not receive growth hormone treatment (table 3).

In order to describe the height of the children and adolescents in reference to the norm for their age and gender the standard deviation score (SDS) of their height was calculated. This represents the deviation of the child's height compared to the average height for their age and gender (see 2.1.1). The children and adolescents participating in the study had an height below -2SD at the beginning of their treatment (see 3.2.2.1), but due to the fact that some of the children received treatment, resulting in a catch-up growth, they had varying degrees of short stature. These SD scores were arranged into three groups in order to describe the degree of short stature. 37.8% of the children and adolescents had a slight deviation from the norm and were grouped in the upper group with a mild degree of short stature. The height of these participants ranged from 0 to -1.49 SD from the norm. 43.7% of the children were categorized into the mid group with a SD from -1.5 to -2.49. The third and lowest group had a standard deviation from -2.5 SD

and below in reference to the norm. This group included 18.5% of all participants (table 3).

SAMPLE CHARACTERISTICS			total	8-12 yrs	13-18 yrs
			N= 137	60	77
Children & adolescents	Age	Ø 13.18			
	Gender	Female	60 (43.8%)	26 (43.3%)	34 (56.7%)
		Male	77 (56.2%)	34 (44.2%)	43 (55.8%)
	Diagnosis	GHD	60 (43.8%)	19 (31.7%)	41 (68.3%)
		ISS	77 (56.2%)	41 (53.2%)	36 (46.8%)
	Treatment status	Treated	83 (60.6%)	31 (37.3%)	52 (62.7%)
		Not treated	54 (39.4%)	29 (53.7%)	25 (46.3%)
	Degree of short stature	Upper group	45 (37.8%)	17 (37.8%)	28 (62.2%)
		Mid group	52 (43.7%)	29 (55.8%)	23 (44.2%)
		Lower group	22 (18.5%)	6 (27.3%)	16 (72.7%)
Parents	Maritial status	Married	92 (76.7%)	42 (45.7%)	50 (54.3%)
		Divorced	16 (13.3%)	4 (25.0%)	12 (75.0%)
		Separated	6 (5.0%)	3 (50.0%)	3 (50.0%)
		Never married	4 (3.3%)	1 (25.0%)	3 (75.0%)
		widowed	2 (1.6%)	1 (50.0%)	1 (50.0%)

Table 3: Sample characteristics

120 participants gave information regarding the marital status of the parents. The majority (76.7%) came from a family where parents were married. 13.3% of the parents were divorced and 5% were separated. 3% of the parents were never married and 1.6% were widowed (table 3).

	SWEDEN	GERMANY	FRANCE	GREAT BRITAIN	SPAIN	TOTAL
GHD	20	16	5	9	10	60
ISS	21	26	15	10	5	77
Total	41	42	20	19	15	137

Table 4: Number of children per country and diagnosis

The sample was a heterogeneous group with respect to the country of origin of the participating children and adolescents. Table 4 shows the distribution of the participants

regarding country and diagnosis. For the retest part of the QOLISSY study the majority of the participants came from Sweden and Germany with together 83 participating children and adolescents.

4.2 Statistical Analysis

In the following part the correlation of the CODI questionnaire and the QOLISSY will be presented, followed by the analysis of both questionnaires regarding the hypotheses presented in the objective.

4.2.1 Correlation CODI und QOLISSY Coping

The Pearson's Correlation of the CODI scales with the total QOLISSY Coping score is shown below in table 21. A very low correlation indicates that there is no connection between these scales. A very high correlation however shows that the scales are very similar. The correlation of the CODI domains with the QOLISSY Coping total score shows a significant correlation on all of the CODI scales, except on the Emotional Reaction scale. The highest small to medium correlation could be seen on the Distance domain. A small correlation could also be seen on the scales Cognitive-Palliative and Avoidance. An even lower correlation was shown on the scales Acceptance and Wishful Thinking. This indicates that both questionnaires do not measure exactly the same concept.

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
QOLISSY Coping total	0.307**	0.326**	0.034	0.257**	0.241**	0.475**

Table 5: Pearson correlation of the CODI domains with the QOLISSY Coping total score

*. Correlation is significant at the 0.05 level (2-tailed).
**. Correlation is significant at the 0.01 level (2-tailed).

In addition to the correlation above a correlation of the CODI scales with the individual QOLISSY Coping items was conducted. Again the CODI domain Emotional Reaction showed little correlation with the QOLISSY Coping items. The Wishful Thinking

scale also showed only a moderate correlation with three of the QOLISSY Coping items and a high correlation with one ("When I feel bad I try to forget about it").

Looking at the items the item "If others tease me I try to stand up for myself" shows only a moderate correlation with the Wishful Thinking and Distancing domain. With the other CODI scales only a slight correlation could be seen. In addition to that the item "When I feel bad about my height I talk about it with my family and or friends" only showed a high correlation with the Cognitive scale and a moderate correlation with the Distance domain (table 22).

Overall the correlation of the two questionnaires only shows mild to moderate correlations. This could be due to the fact that both questionnaires measure not exactly the same concepts. The CODI questionnaire sums up its items to measure six specific coping strategies. The QOLISSY Coping questionnaire however measures individual items. In addition to that the QOLISSY Coping questionnaire puts the focus more on the seeking support strategy, which is not considered directly in the CODI questionnaire.

CODI SCALES						
Items QOLISSY Coping	**Avoid-ance**	**Cogni-tive**	**Emotional Reaction**	**Accept-ance**	**Wishful Think-ing**	**Distance**
I tell myself it is ok to be short	0.193[*]	0.116	-0.120	0.354[**]	0.141	0.381[**]
I try to get used to my height	0.329[**]	0.035	-0.049	0.249[**]	0.189[*]	0.324[**]
If others tease me I stand up for myself	0.083	0.160	0.116	0.077	0.185[*]	0.218[*]
If others tease me my friends stand up for me	0.010	0.059	-0.089	0.261[**]	0.040	0.219[*]
If others tease me I try to talk to them	0.036	0.176[*]	-0.112	0.197[*]	0.036	0.324[**]
When I feel bad about my height I spend time with my friends	0.152	0.279[**]	0.129	0.061	0.077	0.285[**]
When I feel bad about my height I try to think of something nice	0.258[**]	0.272[**]	0.095	0.128	0.211[*]	0.367[**]
When I feel bad about my height I try to think about things I am good at	0.272[**]	0.324[**]	0.063	0.139	0.153	0.342[**]
When I feel bad about my height I talk about it to family and or friends	0.108	0.302[**]	0.052	0.054	0.175	0.182[*]
When I feel bad about my height I try to forget about it	0.450[**]	0.322[**]	0.153	0.076	0.247[**]	0.326[**]

(The leftmost column label for all rows: **QOLISSY Coping items**)

Table 5: Pearson correlation of the CODI scales with the QOLISSY Coping items
*. Correlation is significant at the 0.05 level (2-tailed).   **. Correlation is significant at the 0.01 level (2-tailed).

4.2.2 Coping total and Coping and gender

First the questionnaires were examined regarding the coping strategies and the QOLISSY Coping score in total. Table 5 shows the means and standard deviations of these coping strategies in total. These descriptive statistics show that the sample scores are especially high on the Acceptance scale. In addition to that the scores on Wishful Thinking were relatively high in comparison to the other domains. The lowest scores in total were shown in the Emotional Reaction domain.

SCALE		TOTAL	MALE	FEMALE	T	df	sig.	d
		N = 134	N = 75	N = 59				
Avoidance	M	41.853	37.444	47.458	2.054	132	0.042	0.358
	SD	28.347	28.388	27.519				
Cognitive	M	31.496	32.500	30.254	-0.421	108	0.675	0.071
	SD	32.493	40.178	19.441				
Emotional Reaction	M	18.657	16.444	21.469	1.594	132	0.113	0.277
	SD	18.214	17.516	18.839				
Acceptance	M	70.765	68.457	73.611	0.908	132	0.366	0.154
	SD	32.658	25.463	39.837				
Wishful Thinking	M	58.398	56.164	61.257	0.888	128	0.376	0.158
	SD	32.437	34.219	30.056				
Distance	M	49.532	47.569	52.010	0.179	127	0.417	0.146
	SD	30.725	32.578	28.301				
QOLISSY Coping	M	55.084	55.694	54.360	-0.330	127	0.742	0.058
	SD	22.822	23.610	22.030				

Table 6: Coping scale scores in total and by gender and t-test by gender

In addition to that table 5 contrasts the means and standard deviation of the male and female participants. Comparing the average scores of boys and girls regarding the six coping strategies the only significant difference in scores can be seen in Avoidance (table 6). On the Avoidance scale girls showed on average significantly higher sores (M = 47.46, SD = 27.52) than boys (M = 37.44, SD = 28.39), t(132) = 2.05, p = 0.04, indicating that this coping strategy is more frequently used by girls (table 6).

However, taking the effect size of this differences on the Acceptance scale (d = 0.358) into account it becomes clear that this is only a small to medium effect.

Looking at the QOLISSY coping total score in the sample the mean score was at 55.084 with a standard deviation of 22.822. The gender differences in the score are also illustrated in table 5. In general males scored higher (M = 55.69, SD = 23.61) on the total QOLISSY Coping scale than female participants (M = 54.36, SD = 22.03). These differences were statistically not significant (t(127) = -0.33, p = 0.742) in addition to that, the effect size with d = 0.058 showed no relevant effect.

4.2.3 Coping and age

Table 7 shows the mean and standard deviation of the two age groups on the six CODI coping domains and the QOLISSY total score. In general the children aged 8 to 12 years showed on average higher scores on all the domains, except on the Acceptance scale and the QOLISSY Coping total scale, than the 13 to 18 year old adolescents.

SCALE		8-12 yrs N = 59	13-18 yrs N = 73	T	df	sig.	d
Avoidance	M	48.194	36.711	2.372	132	0.019	0.411
	SD	28.953	26.956				
Cognitive	M	35.085	25.596	1.142	130	0.256	0.284
	SD	41.247	23.053				
Emotional Reaction	M	22.708	15.372	2.358	132	0.020	0.405
	SD	19.878	16.148				
Acceptance	M	64.194	75.933	-2.092	132	0.038	0.352
	SD	41.560	22.397				
Wishful Thinking	M	70.127	48.650	4.052	128	<0.001	0.706
	SD	26.760	33.665				
Distance	M	41.985	55.893	-2.619	127	0.010	0.466
	SD	27.795	31.814				
QOLISSY Coping	M	53.770	56.092	-0.571	127	0.569	0.101
	SD	23.439	22.447				

Table 7: Coping scale scores and t-test by age group

The aspect of the age of the participants showed the most significant differences on the six different Coping scales. Overall on five of the seven coping scales significant differences could be observed between the two age groups. Younger children between

the age of 8 to 12 used the coping strategies of Avoidance significantly more often (M = 48.19, SD = 28.95) than the older children and adolescents between the ages of 13 to 18 (M =36.71, SD = 26.96), t(132) = 2.37, p = 0.019, d = 0.41.

Even though the scores on the Emotional Reaction scale were in general the lowest there were significant differences between the age groups (t(132) = 2.36, p = 0.02, d = 0.41). Younger children had higher scores (M = 22.71, SD = 19.88) indicating a more frequent use of the coping strategy of Emotional Reaction than adolescent participants (M = 15.37, SD = 16.15).

On the Acceptance scale the older age group achieved on average higher scores (M = 75.93, SD = 22.40) indicating that adolescents use the Coping strategy of accepting their short stature significantly more than the younger age group (M = 64.20, SD = 75.93), t(132) = -2.09, p = 0.038, d = 0.35.

Differences between the age groups were also visible regarding Wishful Thinking. This strategy is used far more often in younger children (M = 70.13, SD = 26.76) than in the older age group (M = 48.65, SD = 33.67), t(128) = 4.05, p < 0.001, d = 0.71.

Significant differences also occurred on the Distance scale (t(127) = -2.62, p = 0.01, d = 0.47). The coping strategy Distance is used more often in Adolescents between the ages of 13 to 18 (M = 55.89, SD = 31.81) as in the younger age group (M = 41.99, SD = 27.80).

Looking at the QOLISSY Coping total score adolescents had higher scores (M = 56.09, SD = 22.45) than the younger children (M = 53.77, SD = 23.44), although the differences were not statistically significant (t(127) = -0.571, p = 0.569, d = 0.101).

The observed differences were most distinct on the Wishful Thinking scale, with a calculated effect size of d = 0.706. This indicates a medium to large effect of the age of the participants on the Wishful Thinking domain. The significant differences on the other scales showed medium effects (Distance: d = 0.47; Avoidance: d = 0.41; Emotional Reaction: d = 0.41; Acceptance: d = 0.35) (table 7).

4.2.4 Coping and diagnosis

Table 9 shows the means and standard deviation of the two diagnosis groups on the six CODI scales and the QOLISSY Coping total score.

SCALE		GHD	ISS	T	df	sig.	d
		N = 56	N = 76				
Avoidance	M	42.105	41.667	0.085	104	0.932	0.015
	SD	32.021	25.507				
Cognitive	M	35.089	28.849	1.091	130	0.277	0.182
	SD	43.823	20.471				
Emotional Reaction	M	13.290	22.752	3.237	130	0.002	0.550
	SD	13.506	20.258				
Acceptance	M	78.089	65.175	2.304	132	0.023	0.414
	SD	22.709	37.765				
Wishful Thinking	M	55.324	60.581	-0.910	128	0.365	0.161
	SD	33.907	31.354				
Distance	M	61.690	40.778	4.035	127	<0.001	0.713
	SD	30.907	27.626				
QOLISSY Coping	M	57.057	53.522	0.873	127	0.384	0.153
	SD	25.029	20.959				

Table 8: Coping scale scores and t-test by diagnosis

Significant differences between the children with ISS and GHD were found on the Emotional Reaction scale ($t(130) = 3.24$, $p = 0.002$, $d = 0.55$). Children and adolescents with ISS showed higher scores ($M = 22.75$, $SD = 20.26$) than children with GHD ($M = 13.29$, $SD = 13.50$) (table 9).

Significant differences were also shown regarding the strategies Acceptance ($t(132) = 2.30$, $p = 0.02$, $d = 0.41$) and Distance ($t(127) = 4.04$, $p < 0.001$, $d = 0.71$). In both categories children and adolescents with GHD had higher scores than children with ISS.

The QOLISSY Coping score showed that participants with GHD had higher scores ($M = 57.06$, $SD = 25.03$) than participants with the diagnosis ISS ($M = 53.52$, $SD = 20.96$). However these differences were not statistically significant ($t(127) = 0.87$. $p = 0.384$, $d = 0.153$).

The strongest effects were shown regarding the strategies Distance ($d = 0.71$) and Emotional Reaction ($d = 0.55$).

4.2.5 Coping and treatment status

The table 11 below shows the mean and standard deviation for the group of participants who received treatment and the group who did not receive treatment as well as the t-test and effect sizes.

SCALE		Treated	Untreated	T	df	sig.	d
		N = 83	N = 54				
Avoidance	M	39.896	44.753	-1.014	128	0.312	0.175
	SD	30.603	24.620				
Cognitive	M	31.329	31.745	-0.072	130	0.943	0.013
	SD	38.562	20.718				
Emotional Reaction	M	15.175	23.978	-2.805	132	0.006	0.484
	SD	16.047	20.114				
Acceptance	M	72.418	68.239	0.723	132	0.471	0.121
	SD	24.384	42.454				
Wishful Thinking	M	56.277	61.478	-0.898	128	0.371	0.252
	SD	32.023	33.091				
Distance	M	55.357	40.905	2.683	127	0.008	0.489
	SD	31.686	27.313				
QOLISSY Coping	M	53.568	57.329	-0.918	127	0.361	0.167
	SD	24.193	20.648				

Table 9: Coping scale scores and t-test by treatment status

Comparing the group of the children and adolescents who did not receive treatment with the group who did received treatment showed significant differences on two of the six CODI coping scales. On average untreated children and adolescents scored significantly higher (M = 23.98, SD = 20.11) on the Emotional Reaction scale than the participants who received treatment (M = 15.18, SD = 16.05), t(132) = -2.81, p = 0.006; d = 0.48 (table 12). Considering the effect size a medium effect is shown between these two groups.

In addition to that, children and adolescents who received treatment used the coping strategy of Distancing significantly more (M = 55.36, SD = 31.69) than the untreated participants (M = 40.91, SD = 27.31) with a moderate effect size (t(127) = 2.68, p = 0.008; d = 0.49).

Regarding the QOLISSY total score untreated participants showed higher scores in total (M = 57.33, SD = 20.65) than children and adolescents who did receive treatment (M = 53.57, SD = 24.19), but these differences were not statistically significant (t(127) = -0.92, p = 0.36, d = 0.17).

4.2.6 Coping and degree of short stature

Table 13 shows the means and standard deviation of the three groups of degree of short stature and the results of the ANOVA test, identifying differences in the means of these three groups.

	Upper group		Mid group		Lower group		df	F	sig.
	M	**SD**	**M**	**SD**	**M**	**SD**			
Avoidance	41.861	30.940	37.660	27.440	49.621	22.490	116	1.420	0.246
Cognitive	37.195	49.080	26.538	20.377	36.023	20.969	114	1.330	0.269
Emotional Reaction	15.476	15.840	21.234	19.721	25.568	18.824	115	2.438	0.092
Acceptance	74.008	23.584	67.628	42.712	67.765	24.236	115	0.475	0.623
Wishful Thinking	49.492	32.572	61.583	31.601	68.561	24.658	112	3.184	0.045
Distance	53.125	32.768	45.167	31.984	47.727	22.703	111	0.755	0.472
QOLISSY Coping	51.951	26.514	53.044	21.738	60.357	16.980	111	1.019	0.378

Table 10: Coping scale scores and ANOVA test by degree of short stature

Significant differences could only be seen regarding the Wishful Thinking coping strategy (table 13). Calculating the effect size an effect of d = 0.234 could be seen. In order to investigate between which of the three groups these significant differences occur a post hoc analysis was conducted.

Significant differences were shown on the Wishful Thinking scale between the mid group and upper group with a higher degree of short stature (F(112) = 3.18, p = 0.05). Children and adolescents with a higher degree of short stature had higher scores on the Wishful Thinking scale (M = 68.56, SD = 24.66) than children in the upper group (M = 49.49, SD = 32.57), indicating a more frequent use of this coping strategy.

Another of those trends could be seen on the Emotional Reaction scale. Looking at the means of each of the three group's children and adolescents with a higher degree

of short stature showed higher scores (M = 25.57, SD = 18.82) than the participants in the mid group (M = 21.23, SD = 19.72) and especially than those in the upper group (M = 15.48, SD = 15.84). However, these differences were not statistical significant (p = 0.09).

Examining the QOLISSY Coping total score participants with a mild degree of short stature in the upper group had lower scores (M = 51.95, SD = 26.51) than those in the mid group. Children and adolescents with a higher degree of short stature in the lower group had higher scores (M = 60.36, SD = 16.98) than both of the other groups. These differences were not statistical significant (F(111) = 1.019, p = 0.378). Calculating the effect size though shows a small to medium effect with an effect size of d = 0.377.

4.2.7 Control of the results assuming nonparametric conditions

The results of the tests detailed above were also verified by repeating the tests assuming nonparametric conditions. Due to a slight difference in the distribution across some of the groups the assumptions made for parametric testing could have been compromised. Therefore nonparametric tests were performed, namely the Mann-Whitney-U and the Kruskal Wallis in order to verify the results. Overall the results of the analysis were confirmed. Only slight differences could be seen in the effect sizes. The effect between the diagnosis ISS and GHD on the Acceptance scale increased using nonparametric tests. Still, the effect only increased slightly from a small to medium effect to a medium effect. The detailed results from nonparametric testing can be seen in the Appendix III.

5. Discussion

The thesis presented analyzed data from the retest phase of the international QOLISSY study. The presented results (see chapter 4) will be examined critically and discussed following the hypotheses presented in the in objective (see 3.1). Subsequently a critical reflection will take place and a conclusion and outlook will be shown.

5.1 Discussion of the results

The objective of this thesis was to examine coping mechanisms used in children and adolescents with growth hormone deficiency and idiopathic short stature. Research in this field has been very limited even though coping is considered to be an important aspect in how children and adolescents deal with chronic conditions, in this case short stature. The theoretical framework used in this thesis in which coping is assessed consists of the coping with a chronic diseases model of Meas et al. (1996). This is an extended version of the transactional model of stress and coping of Lazarus (1978) and was adapted to illustrate the coping mechanisms when dealing with a chronic disease. In addition to that, a developmental approach was considered. Many studies show that coping mechanisms and processes are hugely influenced by age and developmental status and changes. It is therefore considered an important aspect in assessing coping mechanisms and strategies.

Looking at the model (figure 1) this thesis examines how different disease and treatment characteristics as well as demographic characteristics influence coping mechanisms and result in different strategies. First it is to say that short stature has a very specific and easily recognizable characteristic to the environment. This usually reflects more problems and results in higher level of coping efforts needed to be applied to deal with the situation. On the other hand short stature is usually not connected with any form of episodes, which children with other diseases have to suffer (e.g. asthma, epilepsy). Therefore the children and adolescents with short stature do not have to fear the loss of controllability and have in this respect a lower level of stress than children suffering from a disease where this is not given.

But there were differences of disease and treatment characteristics even within the sample. The examined children and adolescents could either have idiopathic short

stature or growth hormone deficiency. In addition, treatment characteristics could also potentially influence the coping mechanisms.

The main focus of this thesis was to investigate the differences of coping mechanisms and strategies between different groups of participants. First the coping strategies were examined looking at the whole sample of participants. Overall the strategy Acceptance was used most frequent as well as Wishful Thinking. Emotional Reaction was used least in the sample. Studies show that Wishful Thinking and Emotional Reaction are both strategies often resulting in maladaptive coping processes and representing poorer psychological adjustment (Compas, Connor-Smith, Saltzman, Harding Thompson & Wadsworth, 2001). On the other hand Acceptance is associated with better psychological adjustment (Compas et al., 2001). In order to investigate these different strategies further the sample was examined regarding different variables.

Therefore first gender differences were investigated. The results of the comparison of the two sexes showed significant differences only on the Avoidance scale. Girls showed more Avoidance behavior to deal with problems than boys did. The QOLISSY Coping total score indicated that in total boys had a higher activation of coping than girls, but the differences were not significant. This could be due to the fact that boys are at higher risk for psychosocial problems than girls (Bullinger et al., 2009). According to Lazarus (1966) they are therefore more likely to activate coping mechanisms, because they are faced with more challenges and appraise stressors more likely as harmful. In addition, girls often tend to use Avoidance strategies more than boys, and boys show a higher use of coping than girls (Matud, 2004). However contrary to these literature findings (Tamres et al., 2002; Matud, 2004) no significant differences could be seen on the Emotional Reaction and the Cognitive Palliative scale.

Furthermore age differences were observed in the sample. Age and developmental status are considered to be important factors when assessing coping strategies. The for the thesis relevant developmental stages are middle childhood, represented through the age group 8 to 12 years and adolescents, represented through the age group 13 to 18 years. Characteristics influencing the coping mechanisms and strategies have different impacts on the participants depending on their age. Adolescents are more capable of gaining new resources through mastering developmental tasks and are therefore able to

diversify their coping strategies (Zimmer-Gembeck & Skinner, 2011). Looking at the results regarding differences in age of the participants significant differences could be seen on five of the seven coping scales. Younger children used the coping strategy of Avoidance significantly more than the older group. This finding is conclusive with other research (see 2.2.3) which suggests that younger children use less adaptive and more avoidant strategies than older children (Papastefanous, 2009). Even though the scores on the Emotional Reaction scale were in general the lowest there were significant differences between the age groups. Younger children had higher scores indicating a more frequent use of the coping strategy of Emotional Reaction than adolescent participants. These results are confirmed by other findings which report that younger children have a higher use of Emotional Reaction strategies than older adolescents (Zimmer-Gembeck & Skinner, 2011) Growing up, coping strategies shift away from emotional responses to more adaptive or cognitive responses (Papastefanous, 2009).

On the Acceptance scale the older age group achieved on average higher scores, indicating that adolescents use the Coping strategy of accepting their short stature significantly more than the younger age group. Again this was consistent with other findings (see 2.2.3) reporting a higher use of adaptive strategies such as Acceptance in adolescents than in children Zimmer-Gembeck & Skinner, 2011). Acceptance as a coping strategy is especially used when the stressor is something that has to be adjusted to, because it cannot be changed (Carver, Schleier & Weintraub, 1989). The more frequent use of an Acceptance strategy might therefore due to the child growing up realizing that the short stature is not going to disappear. This argument is also in line with the results regarding Wishful Thinking and Distance.

Wishful Thinking and Distance both represent escapist strategies which research suggests are used more in younger age groups (Zimmer Gembeck & Skinner, 2011). This is mirrored in the results of this thesis. Both strategies are used more often in the children between the ages of 8 and 12 then in the older age group. Younger children have not as developed cognitive abilities than adolescents (Papastefanous, 2009) and therefore might not yet be able to accept their condition as the older age group does. To deal with the stressor they cope with the Wishful thinking strategy which gives temporary relieve from the stressor.

In general the age effects observed in this thesis are in accordance with findings of other studies, however on the QOLISSY questionnaire no significant differences between the two age groups could be observed.

As mentioned above treatment and disease characteristics might also play a role regarding differences in coping mechanisms and strategies. Participants with growth hormone deficit have the potential to catch up with their growth and reach a normal height depending on their treatment status. This option is not available for children and adolescents with idiopathic short stature in Europe. This might be factors influencing the coping with these conditions. Children and adolescents with ISS showed significantly higher use of the coping strategy Emotional Reaction. This might be due to the fact that they are frustrated by the lack of treatment options and try to relieve their frustration by reacting emotionally. Participants with ISS showed also a significantly less frequent use of the strategy Acceptance, whereas children and adolescents with GHD accepted their condition more often. These results indicate a more problematic coping behavior in the group of participants with ISS, as not accepting the condition and reacting emotionally are considered to be associated with poorer psychological functioning. On the other hand children and adolescents with GHD demonstrated a higher use of Distancing behavior, which is also considered to be a strategy associated with maladjustment. Only limited research is available regarding the differences between these two diagnoses; however existing studies suggest no significant differences between these two diagnosis groups (Roberts et al., 2000).

In addition, treatment status is also a characteristic potentially influencing the coping mechanisms. Treated children have the chance to reach a normal height due to the injections with growth hormone, but on the other hand have to deal with the daily injections which some children do not handle well. On average untreated children and adolescents scored significantly higher on the Emotional Reaction scale than the participants who received treatment. Children who are diagnosed with a condition but then do not have a treatment option available might react more emotional than children and adolescents who are presented with a treatment option and therefore a possibility of getting better, resulting in lower scores in this group. In addition to that children and adolescents who received treatment used the coping strategy of Distancing significantly more than the untreated participants. These differences might be due to the already

mentioned daily injections children and adolescents who receive treatment have to cope with. In order not to think about the pain the injections might cause in some cases the children and adolescents try to put the treatment out of their minds. Both strategies are in general associated with poorer psychological adaptation, but in the case of the treated participants the stressor, in this case the short height, is assumed to be temporary. Therefore the Distancing behavior is not as potentially harmful as the strategies applied by the untreated participants. It is only a reaction in order to deal with a temporary stressor. In these cases strategies usually considered to be maladaptive are often en effective way to deal with stress and challenges (Compas et al., 2001; Zimmer Gembeck & Skinner, 2011). Consequently the more problematic coping strategies are applied by the untreated children and adolescents. In addition to that, the overall coping score of the participants who did not receive treatment are higher, even though not statistically significant, indicating a higher use of coping strategies in general. According to Lazarus (1966) coping behavior is only applied if occurring challenges are appraised as potentially harmful. Thus higher use of coping strategies are a result of more problems faced.

Furthermore as another disease characteristic potentially influencing the coping mechanisms differences of coping strategies, the degree of short stature was investigated. Children and adolescents in the lower group with a higher degree of short stature used the coping strategy Wishful Thinking more frequent than children in the upper group with a mild degree of short stature.

Even though the differences on the Emotional Reaction scale were not significant a trend could be seen that children with a higher degree of short stature used the strategy Emotional Reaction more often than those with a milder degree of short stature. This could be due to the fact that children and adolescents with severe short stature face more challenges in their daily life's and therefore have to make a more frequent use of the coping strategies than children and adolescents with a mild form of short stature. This observation was affirmed by the results of the QOLISSY Coping questionnaire. The QOLISSY coping questionnaire showed again no significant differences between the groups. Nevertheless children and adolescents in the lower group seemed to use coping strategies in general more often than participants in the upper group. These findings are in consonance with the coping model illustrated in chapter 2.2.2. According to Lazarus

(1966), a coping response is only triggered when stressors are appraised as potentially harmful. It is to be expected that children and adolescents with a more severe form of short stature are faced with more challenges and therefore need to use coping strategies more often to adapt to and cope with these challenges.

Overall the results of the thesis confirm the hypotheses that there are significant differences in the coping strategies of different sub-groups in the sample of children and adolescents with GHD and ISS. Looking at these differences and the type of coping strategies associated with the different groups assumptions can be made regarding groups with a higher risk of potentially harmful coping strategies. Groups with potentially maladaptive coping behavior are children in the younger age group, participants with the diagnosis idiopathic short stature, who are untreated and are in the lower group with a higher degree of short stature.

5.2 Critical reflection

A number of limitations have to be considered regarding the generalization of the findings.

One aspect that might be influencing the selection of the sample is the contact of the families with the endocrinologic centers. In order to generalize the findings to all children and adolescents with short stature the children and adolescents who contacted the endocrinological centers have to have the same composition and criteria as the children and adolescents who did not. But contacting the clinical centers might be an indicator for individuals with more problems and poorer functioning which would bias the findings.

In addition to that, the willingness to participate in the study might also influence the sample composition. Factors which might be involved in this decision might be the general attitude of the family towards research projects and previous experiences with studies. Other factors might be the impact of the short stature on the life of the family and in general the interest of the family of the condition of their child as well as the personal contact to the clinician.

Furthermore the participants of the study were all aware that they were being studied. This could influence their responses due to different individual motives and interpretation of the study. This awareness of being studied could lead to other

responses than usual and the measure is therefore described as reactive (Sandberg, 2005)

Furthermore there could be a potential of confounding due to the effect of country. The heterogeneity of the sample regarding sample size could restrict the interpretability of the findings.

Overall the results of the CODI questionnaire were compatible with already existing research on the field of coping with chronic diseases. The QOLISSY questionnaire showed in general the same trends, but only few significant differences between the groups could be shown. This could be due to the fact that both concepts do not measure the same concepts, but have slightly different focus. As seen looking at the correlations the QOLISSY questionnaire has an orientation towards assessing support seeking behaviour. The CODI questionnaire on the other hand assesses emotional reactions of the children, which is not assessed explicitly in the QOLISSY questionnaire. Furthermore, the few significant differences could also be due to the fact, that the QOLISSY questionnaire is disease specific for short stature, whereas the CODI questionnaire is a chronic generic instrument, measuring on the level of chronic diseases.

These limitations have to be considered when trying to generalize the findings of this study. However taken this into account the results can serve as an indicator for future studies into the field of coping strategies in short stature youth and potentially important factors to be considered.

6. Conclusion and Outlook

In this thesis an attempt was made to assess the different coping mechanisms and strategies applied by children and adolescents with growth hormone deficiency and idiopathic short stature. Coping with short stature is an important aspect in the life of children and adolescents with GHD or ISS. Identifying the different coping strategies used by these children could be seen as an important tool to improve the ways of coping with problems and challenges faced by children with short stature every day. It allows to identify differences in coping strategies in different groups and therefore helps for example to identify groups with increased use of maladaptive coping strategies. The results show that especially young children with ISS, who do not receive any treatment use potentially maladaptive coping strategies. This demonstrates the need for assistance for these children and adolescents. Especially the influence of age has to be recognised in approaches to improve the use of coping strategies. The development during childhood and adolescents seems to play an important part in the coping process. Therefore further studies examining coping during childhood and adolescents have to take these differences into account.

In addition to that growing up with short stature presents a special challenge to children and adolescents as well as their parents. The treatment option for these conditions are limited, hence efforts are being made to provide the children and adolescents with psychosocial support and interventions developed to improve the quality of life in short stature youth. This is also considered as a possible alternative to GH treatment in light of the fact that children and adolescents with idiopathic short stature are not viable for GH treatment in Europe. However interventions trying to tackle these problems should be designed with a clear developmental approach and support children and adolescents appropriate due to their age and developmental stage.

The QOLISSY and CODI questionnaires can not only be used as an outcome measurement in clinical studies, but can also be used to assess needs of individual patients and groups of patients. This could be used as a point of action to develop an intervention. This intervention would ideally close the gap between medical treatment for

GHD patients and lack of treatment for ISS patients. In this aspect it would be especially interesting to see the influence of coping on quality of life in short stature youth.

This intervention has to be based on patients needs as assessed in prior project and reaffirmed in this thesis. Especially children and adolescents with ISS suffer. They often receive the diagnosis, but are left without any treatment options. This is not a satisfactory situation for the children or the parents who often experienced enormous difficulties leading them to consult a physician in the first place. In order to investigate if psychological counselling combined with or in place of GH treatment is beneficial an approach has to consist of a carefully planned and documented intervention program. To select participants for the intervention a quality of life screening with questionnaires for example the QOLISSY questionnaire (Quitmann et al., 2010), and instruments to assess psychosocial variables and coping.

The intervention might consist of modularized sessions each dealing with a specific topic of concern. This could be administered in a few sessions in which the resources and strains on the life of the children and adolescents are assessed and techniques to cope with these difficulties are practiced. This intervention should have a strong focus on coping with challenges and problems and learning how to deal with them considering their different developmental phases. It is also important to include the parents in these interventions, as they also have a tremendous influence on the health and wellbeing of their children (Kliewer & Lewis, 1995).

In addition to that it would be interesting to recruit a control group to assess the coping strategies of children without short stature and hence have a basis to compare the different strategies used in children and adolescents with and without short stature.

In conclusion, children and adolescents with GHD and ISS use various coping strategies, some of them maladaptive and contra productive to the health and wellbeing of the children and adolescents. Especially children with idiopathic short stature, without any treatment are affected by these strategies. Psychosocial support groups would enable children to identify, understand and manage the demands in life of having short stature by applying balanced coping strategies (Creedy et al., 2004)

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Appendix

Appendix I: The CODI questionnaire

CODI

Think of situations, when you have been **bothered or stressed** because of your illness. Below you find a list of things how kids may deal with their illness in these situations.

Please tell us, **how often** you usually do the things or have this kind of thoughts related to your illness.

Avoidance		never	seldom	quite often	very often	always
1.	... I try to forget my illness.	❑	❑	❑	❑	❑
2.	... I pretend to be all right.	❑	❑	❑	❑	❑
3.	... I try to ignore my illness.	❑	❑	❑	❑	❑

Cognitive Palliative		never	seldom	quite often	very often	always
4.	... I believe that faith in god helps me.	❑	❑	❑	❑	❑
5.	... I pray that my illness will go away.	❑	❑	❑	❑	❑
6.	... I learn as much as possible about my illness.	❑	❑	❑	❑	❑
7.	... I tell myself that even famous people have illnesses.	❑	❑	❑	❑	❑
8.	... I think of worse situations.	❑	❑	❑	❑	❑

Emotional Reaction		never	seldom	quite often	very often	always
9.	... I am angry.	❑	❑	❑	❑	❑
10.	... I cry.	❑	❑	❑	❑	❑
11.	... I am frustrated.	❑	❑	❑	❑	❑
12.	... I am ashamed of being ill.	❑	❑	❑	❑	❑
13.	... I think it is unfair that I am ill.	❑	❑	❑	❑	❑
14.	... I wake up at night and think of terrible things.	❑	❑	❑	❑	❑

Acceptance		never	seldom	quite often	very often	always
1.	... I accept my illness.	❑	❑	❑	❑	❑
2.	... I got used to my illness.	❑	❑	❑	❑	❑
3.	... I am able to manage my illness.	❑	❑	❑	❑	❑
4.	... I cope well with my illness.	❑	❑	❑	❑	❑
5.	... I face my situation with humour.	❑	❑	❑	❑	❑
6.	... I take my illness easy.	❑	❑	❑	❑	❑

Wishful Thinking		never	seldom	quite often	very often	always
7.	... I hope that my illness disappears.	❑	❑	❑	❑	❑
8.	... I want to stop having my illness.	❑	❑	❑	❑	❑
9.	... I wish I was healthy.	❑	❑	❑	❑	❑

Distance		never	seldom	quite often	very often	always
10.	... I think my illness is not so serious.	❑	❑	❑	❑	❑
11.	... I don't care about my illness.	❑	❑	❑	❑	❑
12.	... I think my illness is no big deal.	❑	❑	❑	❑	❑
13.	... I forget about my illness.	❑	❑	❑	❑	❑

Overall, how well do you think you cope with your illness?		1	2	3	4	5
14.	1=not well at all 5= very well	❑	❑	❑	❑	❑

Appendix II: The QOLISSY questionnaire

🧑QoLISSY🧑 C

Date: _____ Name: _____

Date of Birth: _____ First name: _____ ID:_____

Hi!

We are interested in how you feel about yourself and we would like to invite you to help us find out. The questions relate to your life in general, your height and your strengths and difficulties.

- Please answer the following questions. There are no right or wrong answers: the most important thing is that you tell us HOW *YOU* FEEL. It is important that you answer ALL the questions and also that we can see your marks clearly.

- When you think of your answer please remember the past week. If a question is difficult to answer please try it nevertheless by choosing the closest answer – you can also write a note on the last page.

- You do not have to show your answers to anybody else except us. Also, nobody who knows you will look at your questionnaire once you have finished it. And we will not keep your name together with your questionnaire.

- If you have any difficulties or concerns please write them down on the last page.

This is how it works:

Please read every sentence below carefully. What answer comes to your mind first? Choose the box that fits your answer best and mark it with an X.

Sometimes we would like to know **HOW STRONGLY** you think or feel something, and sometimes **HOW OFTEN** you think or feel something. When you think of your answer please try to remember the last week, meaning the last seven days.

Example

Thinking about last week:	Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
I like pizza?	○	○	○	○	⊗

If you love eating pizza, then you would mark the box that says "Extremely/always".

		Let us begin with problems you might have with your height.				
1.1	My height prevents me from doing things that other children my age do.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○
1.2	Because of my height I have problems everyday.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○
1.3	Because of my height I have more trouble reaching things than others my age.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○
1.4	Because of my height I depend on others.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○
1.5	I have to look up at others my age when I talk to them.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○
1.6	It bothers me that others my age can go on fairground rides and I can't.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely / always ○

		In this part we would like to know what it is like for you to be with other people (e.g. your family, friends, classmates, strangers).				
2.1	Being asked about my height at school bothers me.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.2	I feel small around others my age.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○

2.3	Others mistake me for being younger than I am.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.4	Because of my height I get laughed at or teased.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.5	Because of my height I am treated differently.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.6	My height is the only thing others notice about me.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.7	Because of my height I have problems getting the clothes I like.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
2.8	It hurts to be left out of things because of my height.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○

This part is about your emotions and how you feel about your height.

3.1	Because of my height I feel different from others my age.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
3.2	I am fed up with comments about my height.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
3.3	Because of my height I am shy.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○
3.4	I am happy with my height.	Not at all/never ○	Slightly/ seldom ○	Moderately/ quite often ○	Very/very often ○	Extremely/ always ○

3.5	I am insecure because of my height.	Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
		O	O	O	O	O
3.6	I am sad because of my height.	Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
		O	O	O	O	O
3.7	Despite my height, I feel comfortable with the way I am.	Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
		O	O	O	O	O
3.8	My height bothers me.	Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
		O	O	O	O	O

Sometimes things might not be easy for you. Please tell what you think or do to feel better.

|						
4.1	I tell myself it is ok to be short.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.2	I try to get used to my height.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.3	If others tease me I stand up for myself.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.4	If others tease me my friends stand up for me.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.5	If others tease me I try to talk to them.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.6	When I feel bad about my height I spend time with my friends.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.7	When I feel bad about my height I try to think of something nice.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.8	When I feel bad about my height I try to think about things I am good at.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.9	When I feel bad about my height I talk about it to family and or friends.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
4.10	When I feel bad about my height I try to forget about it.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯

What do you think about height in general?

		Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
5.1	I believe that being taller would make me happier.	O	O	O	O	O
5.2	I believe that tall people are better liked.	O	O	O	O	O
5.3	I believe that it is important to be tall.	O	O	O	O	O
5.4	I believe that being tall helps in life.	O	O	O	O	O

Please answer the following questions only if you receive or if you have received growth hormone therapy.

		Not at all/never	Slightly/ seldom	Moderately/ quite often	Very/very often	Extremely/ always
6.1	Because of my treatment I am stronger and more energetic.	O	O	O	O	O
6.2	My friends understand that I need treatment.	O	O	O	O	O
6.3	I feel good because of my treatment.	O	O	O	O	O
6.4	My treatment really helps me to grow.	O	O	O	O	O
6.5	Taking my injections every day is annoying.	O	O	O	O	O
6.6	The injections are painful.	O	O	O	O	O
6.7	Because of the treatment I am physically stronger.	O	O	O	O	O
6.8	Because of the treatment I am happier about myself.	O	O	O	O	O
6.9	Because of the treatment I can join in with my friends.	O	O	O	O	O

6.10	Because of the treatment I am able to concentrate better.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
6.11	Because of the treatment, it is now easier to reach things.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
6.12	Because of the treatment, I am better at sports.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
6.13	Because of the treatment, I can wear the clothes I like.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
6.14	I feel more comfortable in the presence of others.	Not at all/never ◯	Slightly/ seldom ◯	Moderately/ quite often ◯	Very/very often ◯	Extremely/ always ◯
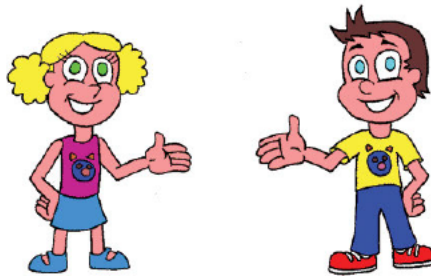
If you want to write about something that was not in the questionnaire, please do that here.

Thank you very much!

Appendix III: Tables of nonparametric tests

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
Mann-Whitney-U	1749.000	2025.500	1833.500	2102.000	1948.000	1858.500
Z	-2.089	-0.588	-1.714	-0.530	-0.625	-0.920
Significance	0.037	0.557	0.087	0.596	0.532	0.357
r	-0.180	-0.051	-0.148	-0.046	-0.055	-0.081

Table 11: Mann-Whitney-U test by gender and effect size

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
Mann-Whitney-U	1710.000	1939.500	1722.000	1477.000	1338.000	1545.500
Z	-2.295	-0.983	-2.248	-3.310	-3.557	-2.463
Significance	0.022	0.326	0.025	<0.001	<0.001	0.014
r	-0.198	-0.085	-0.194	-0.286	-0.312	-0.217

Table 12: Mann-Whitney-U test by age group and effect size

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
Mann-Whitney-U	2165.000	2086.000	1620.000	1402.500	1885.500	1254.500
Z	-0.133	-0.194	-2.646	-3.614	-0.791	-3.689
Significance	0.894	0.846	0.008	0.000	0.429	0.000
r	-0.012	-0.017	-0.229	-0.312	-0.069	-0.324

Table 13: Mann-Whitney-U test by diagnosis, and effect size

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
Mann-Whitney-U	1911.500	1841.000	1571.500	1785.000	1833.500	1470.500
Z	-1.133	-1.176	-2.640	-1.651	-0.986	-2.559
Significance	0.257	0.240	0.008	0.099	0.324	0.010
r	-0.097	-0.102	-0.228	-0.143	-0.086	-0.225

Table 14: Mann-Whitney-U test by treatment status, and effect size

	Avoidance	Cognitive	Emotional Reaction	Acceptance	Wishful Thinking	Distance
Chi-Squared	2.916	3.609	4.965	3.280	5.569	0.960
df	2	2	2	2	2	2
Significance	0.233	0.165	0.084	0.194	0.062	0.619

Table 15: Kruskal Wallis test on the CODI scales

QOLISSY COPING		Mean rank	Mann-Whitney-U	Z	sig.	r
Gender	*male*	65.91	2001.000	-0.300	0.764	-0.028
	female	63.92				
Age group	*8-12*	62.26	1890.500	-0.730	0.465	-0.064
	13-18	67.10				
Diagnosis	*GHD*	68.95	1827.000	-1.068	0.286	-0.094
	ISS	61.88				
Treatment status	*treated*	62.97	1846.000	-0.750	0.454	-0.070
	untreated	68.00				
			Chi2	df	sig	
Severity of short stature	*mild*	53.72	2.074	2	0.355	
	moderate	54.95				
	severe	65.62				

Table 16: Total QOLISSY Coping score by various variables

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