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# The role of acceptance and goal regulation in well-being of adolescents with cystic fibrosis and diabetes

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# GENERAL INTRODUCTION

Chronic illnesses can be defined as illnesses with a prolonged course that do not resolve spontaneously, for which a complete cure is seldom achieved, and that cause functional impairment or disability (Brownson, Remington, & Davis, 1998). Two chronic illnesses showing an upward trend are cystic fibrosis (due to increased rates of survival into adulthood) and diabetes mellitus (due to a growing incidence) (Cystic Fibrosis Foundation, 2010; International Diabetes Federation, 2006). Both illnesses affect several organs throughout the body (e.g., lungs and pancreas for cystic fibrosis, heart and kidneys for diabetes) what leads to a complex and invalidating clinical syndrome and a shortened life span (Davis, 2006; International Diabetes Federation, 2006). The limiting character of both diseases forces affected individuals into a complex and intensive daily treatment regimen. For adolescents, this deteriorating health and demanding treatment may seriously impact upon their general well-being (Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Yeo & Sawyer, 2005). Therefore, it is essential to understand how adolescents with chronic illness can maintain their well-being despite the burden of being ill.

## CYSTIC FIBROSIS

Cystic fibrosis is the most common life-shortening genetic condition amongst the Caucasian population, affecting 1 in 2500 newborn children (Cystic Fibrosis Worldwide, 2005; Ratjen & Döring, 2003). Basic defect is a mutation of the CFTR gene, which disturbs the transport of salt and water in and out the body cells. This results in an increased viscosity of the exocrine secretions throughout the body (Kerem et al., 1989; Quinton, 1983; Riordan, 1989; Rommens et al., 1989). These hyper viscous secretions mainly affect the lungs and the pancreas by plugging the airways and pancreatic ducts. Plugging causes recurrent cycles of infection and inflammation and/or progressive and irreversible tissue damage (Ratjen & Döring, 2003; Rosenstein & Zeitlin, 1998).

In the lungs, infection, inflammation, and tissue damage cause several symptoms such as a chronic productive cough, shortness of breath, wheezing, pain, chronic sinusitis, and nasal polyps (Rosenstein & Zeitlin, 1998). Long-term complications are coughing blood, pneumothorax, hypoxemia, respiratory insufficiency, cor pulmonale, and heart failure. Lung damage in cystic fibrosis is slow but progressive (Davies, Alton, & Bush, 2007). At

a certain point there is insufficient functional lung tissue to provide the body with oxygen, with death as a consequence. Lung destruction is responsible for more than 95% of all cystic fibrosis-related deaths (Davis, 2006).

Obstruction in the pancreas leads to pancreatic insufficiency due to autodigestion of the pancreatic ducts. This manifests itself in a defective digestion and absorption of nutrients with symptoms such as abnormal stool, malnutrition, abdominal pain, growth and pubertal delay (Davies et al., 2007; Rosenstein & Zeitlin, 1998). Long-term complications are reflux, rectal prolapse, bowel obstruction, diabetes, and liver disorders (Davies et al., 2007; Ratjen & Döring, 2003).

Besides lung and pancreatic damage, also other organs are affected. Frequently occurring complications are osteopenia, arthritis, and clubbing of the fingers (Davies et al., 2007). Furthermore, both males and females can be affected by infertility (Davies et al., 2007; Edenborough, 2001; Patrizio & Salameh, 1998; Rosenstein & Zeitlin, 1998). In males, 98% is infertile, but not sterile, due to absence of the vas deferens (Patrizio & Salameh, 1998). In females fertility problems are much less frequent but can occur due to thick cervical mucus, malnutrition, and amenorrhea (Edenborough, 2001).

Up till now, there is no curative treatment for cystic fibrosis. Treatment is focused on the suppression of symptoms, the prevention of damage to the organs concerned, and the increase of quality of life. Daily treatment consists of a high calorie diet, medicines (e.g., anti-inflammatory drugs, pancreatic enzymes, mucolytica, antibiotics), exercise, and airway clearance (e.g., chest physical therapy, vibration). Furthermore, routine hospital visits and sometimes hospitalisation are necessary (Davies et al., 2007). Thanks to significant advances in medical care, the current median age of survival for individuals with cystic fibrosis runs up to 37.5 years (Cystic Fibrosis Foundation, 2010).

**DIABETES MELLITUS**

Diabetes mellitus is a group of chronic medical conditions characterised by hyperglycaemia (i.e., raised blood sugar) and intolerance of glucose (Harris & Zimmet, 1997; World Health Organisation, 2011a). Type 1 diabetes is one of the most common metabolic conditions in children and adolescents and its prevalence is still increasing (Patterson et al., 2009). Type 2 diabetes used to be rare in children and adolescents, but now counts for approximately 50% of all new diabetes diagnoses in children and adolescents worldwide (Fagot-Campagna et al., 2001; World Health Organisation, 2006).

Basic defects are the autoimmune destruction of β-cells in the pancreas, leading to defective insulin secretion (type 1 diabetes) or deficient tissue responses to insulin leading to defective insulin action together with a inadequate compensatory insulin secretion reaction (type 2 diabetes) (Harris & Zimmet, 1997; The Expert Committee on the diagnosis and Classification of Diabetes Mellitus, 2002; World Health Organisation, 2011b). Causes of childhood type 1 diabetes are still being investigated, but seem to include genetic predisposition, gestational factors (e.g., congenital rubella, birth order, maternal age, high birth weight) and environmental factors (e.g., stress events, nutrition) (Larsson et al., 2004). Risk factors for type 2 diabetes comprise high body weight, physical inactivity, insulin resistance, and genetic predisposition (International Diabetes Federation, 2006; World Health Organisation, 2011b).

Hyperglycaemia leads to cardiovascular diseases such as stroke, myocardial infarction, angina pectoris, peripheral artery disease, and congestive heart failure which are responsible for 50% and more of all diabetes-related deaths (International Diabetes Federation, 2006; Wilson, 1998; World Health Organisation, 2011b). In the kidneys, diabetes may lead to structural (e.g., increased renal size) and functional changes (e.g., loss of renal function) with symptoms of kidney dysfunction such as fatigue, headache, nausea, poor appetite, and swelling of the lower limbs (Gellman et al., 1959; International Diabetes Federation, 2006; Mauer et al., 1984; Mogensen, 1976; Viberti, Pickup, Jarrett, & Keen, 1979). Nephropathy is a major cause of morbidity and mortality in individuals with diabetes and may lead to the need for dialysis or kidney transplantation (Gall, Borch-Johnson, Hougaard, Nielsen, & Parving, 1995; MacLeod, Lutale, & Marshall, 1995; Mogensen, 1984; Rossing, Hougaard, Borch-Johnson, & Parving, 1996; Valmadrid, Klein, Moss, & Klein, 2000).

Diabetes also commonly leads to damage of the nerves of the peripheral nervous system (i.e., neuropathy). This damage is manifested by disturbances in several innervated organ systems such as the cardiovascular, gastrointestinal, and ocular systems (Vinik, Raelene, Braxton, & Freeman, 2003). Typical symptoms only occur in the long run and comprise hearts rate disturbances, postural hypotension, exercise intolerance, silent myocardial ischemia, vomiting, nausea, malabsorption, diarrhoea, constipation, incontinence, excessive sweating, loss of feeling in the foot, foot ulceration (and subsequent lower limb amputation), bladder dysfunction, urinary retention, sexual dysfunction, and pain (Ewing & Clarke, 1982; Vinik et al., 2003).

Another complication of diabetes is retinopathy, a retinal vascular disorder. Diabetic retinopathy is characterised by microaneurysms, intraretinal hemorraghes, proliferation of new but fragile blood vessels in the retina, and intrusion of these vessels in the vitreous cavity, the iris anterior chamber of the eye (Aiello et al., 1998; Frank, 2004; Henkind, 1978; Moss, Klein, & Klein, 1988; Patz, 1982). Over time, this leads to visual loss (and sometimes blindness), neovascular glaucoma, and excessive intraocular pressure (Zhang et al., 2010).

Today there is no cure for diabetes but effective treatment to reduce the risk of complications exists. Main goal of this treatment is to maintain glycemic levels close to the nondiabetic range by a strict regimen that may include monitoring of complications, self-monitoring of blood glucose, insulin and/or pharmacological therapy, and/or life style intervention (i.e., weight loss/healthy eating and increased physical activity) (International Diabetes Federation, 2011; Nathan et al., 2006, 2009). Despite this treatment, diabetes reduces life expectancy up to 10 years for individuals with type 2 diabetes and more than 20 years in individuals with type 1 diabetes (Diabetes UK, 2010).

**ACCEPTANCE AND WELL-BEING**

As previous research has indicated that chronic illness may seriously impact upon the well-being of affected adolescents, it is important to determine how their well-being can be maximized and sustained. Generally, it has been suggested that the way of evaluating illness may explain individual differences in well-being (Evers, et al., 2001; Leventhal, Brisette, & Leventhal, 2003; Maes & Karoly, 2005). An important and adaptive type of evaluation of illness is 'acceptance'. Acceptance can be defined as ''recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences'' (Evers et al., 2001, p. 1027). Research indeed has shown that acceptance may play a protective role in the well-being of children and adolescents with chronic illnesses such as chronic pain (e.g., McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011), juvenile arthritis (Feinstein et al., 2011), sickle cell disease (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011). Specifically, this research has shown that more acceptance is related to positive outcomes such as less anxiety, less depression, less disability, and better emotional, social, and physical functioning.

Next to investigating if acceptance of illness explains differences in well-being in adolescents with chronic illness, it is also important to gain insight in the processes

through which acceptance may affect well-being. Identifying these processes is essential to develop or improve interventions that aim at maximizing the well-being of these adolescents. The current dissertation focuses on two specific goal regulation processes (i.e., goal interference and goal facilitation) as potential underlying processes. Reasons for this focus are that acceptance is theoretically assumed to play a key role in the regulation of one's personal goals, and that goal regulation has increasingly been found to explain well-being in the context of chronic illness (De Ridder & Kuijer, 2007; Gebhardt, 2008; Maes & Karoly, 2005).

## GOAL REGULATION

Human life can be described as a continuous regulation of personal goals (Carver & Scheier, 1998). Goal regulation, across all theories, can be defined as the process of setting goals and tuning behaviour to the attainment of these goals (Maes & Karoly, 2005). Goals as such can generally be defined as internal representations of future states one tries to attain, maintain, or avoid (Austin & Vancouver, 1996; Carver & Scheier, 1998). All goal theories endorse the idea that goals stimulate and organize behaviour thereby providing meaning and direction to life (Carver & Scheier, 1998). Indeed, extensive research has shown that setting and attaining valuable life goals plays a key role in well-being (Austin & Vancouver, 1996; Carver & Scheier, 1998; Deci & Ryan, 2000; Emmons, 1986; Little, 1993).

### Goal regulation in adolescence

During adolescence, goals are especially important as they reflect age-specific life tasks that need to be achieved in the transition to adulthood (Cantor, Norem, Niedenthal, Langston, & Brower, 1987; Erikson, 1963; Massey, Gebhardt, & Garnefski, 2008; Nurmi, 1987, 1991). By setting and pursuing goals adolescents steer their own development and form an own identity (Erikson, 1963; Nurmi, 1991, 1993, 2001). Conversely, this development and identity also steers the setting and pursuit of goal, creating a continuous reciprocal relationship (Nurmi, 2001). The goals unique for adolescence relate to the rapid and extensive physical (e.g., sexual maturation), cognitive (e.g., increased abstract reasoning), social (e.g., developing stable relationships), and emotional growth (e.g., emotional independence, changing emotional reactions) characteristic for this period (Ausubel, 2002; Massey, Gebhardt, & Garnefski, 2008).

There are no absolute criteria to mark the adolescence period in time. In general, the start of adolescence is defined by biology (e.g., physical maturation), while the end is

defined by the integration of the individual in adult society (Ausubel, 2002). Adolescence thus can be considered as a sustained period of personal development where goal regulation is intensively trained and refined (Massey, 2009; McGrath, 1999).

**Goal setting**

Adolescence is characterised by an increasing autonomy in the setting of goals (Nurmi, 1993). Goals that are particularly important during this period relate to education, occupation, identity, and social relationships (Cross & Markus, 1991; Little, 1993; Nurmi, 1992; Massey et al., 2008; Salmela-Aro & Nurmi, 1997). The type of goals adolescents set depends on several factors, such as life events, life history, prior goal striving, personal factors (e.g., gender, age), social factors (e.g., influence of others), normative factors (e.g., institutional tracks), and biological factors (e.g., illness, disability) (Deci & Ryan, 2000; Emmons, Colby, & Kaiser, 1998; Leventhal, Brisette, & Leventhal, 2003; Leventhal, Halm, Horowitz, Leventhal, & Ozakinci, 2004; Maes & Karoly, 2005; Nurmi, Poole, & Seginer, 1995; Salmela-Aro & Nurmi, 1997). Specifically, illness like cystic fibrosis and diabetes creates the need to set new goals in response to this illness (Badlan, 2006; Dijkstra, Buunk, Tóth, & Jager, 2008; Emmons et al., 1998; Seiffge-Krenke, 1998; Zeltzer, 1985). These goals are crucial for managing, adapting and giving meaning to illness (De Ridder & Kuijer, 2007; Schwartz & Drotar, 2006). Illness-related goals include, for example, engaging in treatment, establishing a good relationship with medical personnel, and taking responsibility for treatment. However, despite the (even vital) importance of illness-related goals, they often do not get prioritized attention (cfr. low treatment adherence rates in chronic illness) (Gebhardt, 2006; 2008).

**Goal pursuit and attainment**

Individuals not always succeed in attaining their personal goals. Several factors such as life events (e.g., parental divorce), personal resources (e.g., time and energy), socio-structural factors (e.g., expected developmental transitions), biological factors (e.g., illness), normative factors (e.g., social norms), and the presence of other goals can facilitate or hinder one's goal attainment (Brandstädter & Renner, 1990; Emmons et al., 1998; Wrosch & Freund, 2001). Especially chronic illness can strongly influence one's goal attainment (Rothermund, 2006; Schwartz & Drotar, 2006). First, illness may limit the attainment of personal goals because of the symptoms, physical disability, and worsening health it entails (Badlan, 2006; Dijkstra, Buunk, Tóth, & Jager, 2008; Emmons et al., 1998; Schwartz & Drotar, 2009; Seiffge-Krenke, 1998; Zeltzer, 1985). Second,

illness may create new goals (e.g., treatment). As goals are not necessarily independent, striving for treatment goals may interfere with other personal goals (Badlan, 2006; Davis, 2006; Dijkstra et al., 2008; Emmons & King, 1988; Little, 1983; Schwartz & Drotar, 2006). Goal interference occurs when striving for one goal decreases the chance of attaining another goal (Little, 1983; Riediger, 2001; Riediger & Freund, 2004). This can be due to the fact that resources for goal striving (e.g., time, energy) are limited and/or that the strategies for attaining both goals are incompatible (Riediger, 2001; Riediger & Freund, 2004). Previous research has shown that goal interference is related to less well-being (Riediger & Freund, 2004). Third, the new goals created by illness may also facilitate other personal goals. Goal facilitation occurs when striving for one goal increases the chance of attaining another goal (Little, 1983; Riediger, 2001; Riediger & Freund, 2004). Facilitation can be due to instrumental relations between goals and/or compatible strategies for attaining these goals (Riediger, 2001). Goal facilitation and interference are conceptualised as independent characteristics. This means that goals may facilitate each other in one way, and, at the same time, may interfere with each other in another way (Little, 1983; Riediger & Freund, 2004). For example, the goal 'engaging in treatment' may facilitate the goal 'getting good grades' as it contributes to better health. At the same 'engaging in treatment' may interfere with 'getting good grades' as it takes a lot of time, leaving less time to study. It is clear that chronic illness may seriously impact upon goal regulation and related well-being of affected individuals. Therefore it is necessary to investigate how successful regulation of illness-related and other personal goals and well-being can be achieved despite being ill.

## SUMMARY

In chronic illness, the permanent health threat and demanding treatment may seriously impact upon the well-being of affected individuals (Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Schwartz & Drotar, 2006, 2009; Yeo & Sawyer, 2005). Based on previous research in several chronic illnesses, 'acceptance of illness' can be identified as a process that has the potential to influence well-being in adolescents with chronic illness (e.g., Evers et al., 2001; McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011). Next to investigating the potential protective role of acceptance of illness in well-being, understanding of the processes through which acceptance may affect well-being is essential. The current set of studies focusses on goal interference and goal facilitation as possible mediating processes. These processes are

especially important for adolescents with chronic illness as during adolescence individuals need to achieve important life tasks to pass on to adulthood, start to regulate their goals more autonomously, are cognitively capable to evaluate the impact of illness on goal regulation, have a strong desire to be 'normal', tend to be non-adherent to treatment, and because illness creates the need to pursue treatment goals, what may interfere with the pursuit of other life goals (e.g., becoming independent) (Badlan, 2006; Little, 1983; Massey, et al. 2008; Nurmi, 1987, 1991,1993; Schwartz & Drotar, 2006, 2009).This dissertation describes six studies each investigating a combination of these objectives in various samples and by means of different methodologies.

**OUTLINE OF DISSERTATION**

The current dissertation aims to investigate (1) the role of acceptance of illness in well-being of adolescents with chronic illness, (2) the role of acceptance in the goal regulation of these adolescents, (3) the mediating effect of goal interference and facilitation on the relationship between acceptance and well-being, and (4) differences in goal regulation between adolescents with and without chronic illness. Well-being is operationalised in terms of positive affect, negative affect, and life satisfaction (Diener, 1984; Diener, Suh, Lucas, & Smith, 1999). Positive affect refers to moods and emotions such as happiness, joy, and contentment whereas negative affect comprises emotional responses such as anxiety, depression, and stress (Diener et al., 1999). Life satisfaction, as third component, includes a global evaluation of one's life across different domains (Diener, 1984; Diener et al., 1999).

**Chapter 1** describes a cross-sectional questionnaire study, in which the role of acceptance in negative affect of adolescents with cystic fibrosis ($N = 34$, 12-22 years) was investigated. Specifically, this study focused on the role of acceptance in anxiety, depression, and functional disability.

**Chapter 2** elaborates these findings by performing a prospective analysis of the role of acceptance in negative affect and life satisfaction in adolescents with cystic fibrosis (Time 1: $N = 40$, Time 2: $N = 28$; 14-22 years). By means of questionnaires, the relationship between acceptance (measured at Time 1) and anxiety, depression, and quality of life (measured at Time 1 and 2) was examined.

**Chapter 3** once more examined the role of acceptance in negative affect of adolescents with cystic fibrosis ($N = 30$; 12-22 years) but his time under circumstances specific for chronic illness (i.e., during medical procedures). This study investigated the

role of acceptance in self-reported distress (i.e., anxiety/worry about the procedure), physiological distress (i.e., heart rate and heart rate variability), expected pain and pain-related thoughts during spirometry. Acceptance, self-reported distress, expected pain and pain-related thought were assessed by questionnaires, while physiological distress was measured by a heart rate monitor.

In **chapter 4** goal regulation is introduced. Goals and goal regulation processes were compared in a questionnaire study in adolescents with ($N = 60$; 14-22 years) and without chronic illness ($N = 60$; 14-22 years).

**Chapter 5** analyses the mediating of goal interference and facilitation in the relationship between acceptance and well-being in adolescents with cystic fibrosis and diabetes ($N = 30$; 14-22 years) on a daily basis.

**Chapter 6** describes a questionnaire study in adolescents with cystic fibrosis and diabetes ($N = 38$; 14-22 years), investigating the mediating role of goal interference and facilitation in the relationship between acceptance and well-being. A prospective design was used: acceptance, goal interference and facilitation, and well-being were assessed at three moments in time (0 months, 2 months, 4 months).

The **general discussion** highlights the main results of the different studies. Furthermore, clinical and theoretical implications, limitations of the current studies and ideas for future research are discussed.
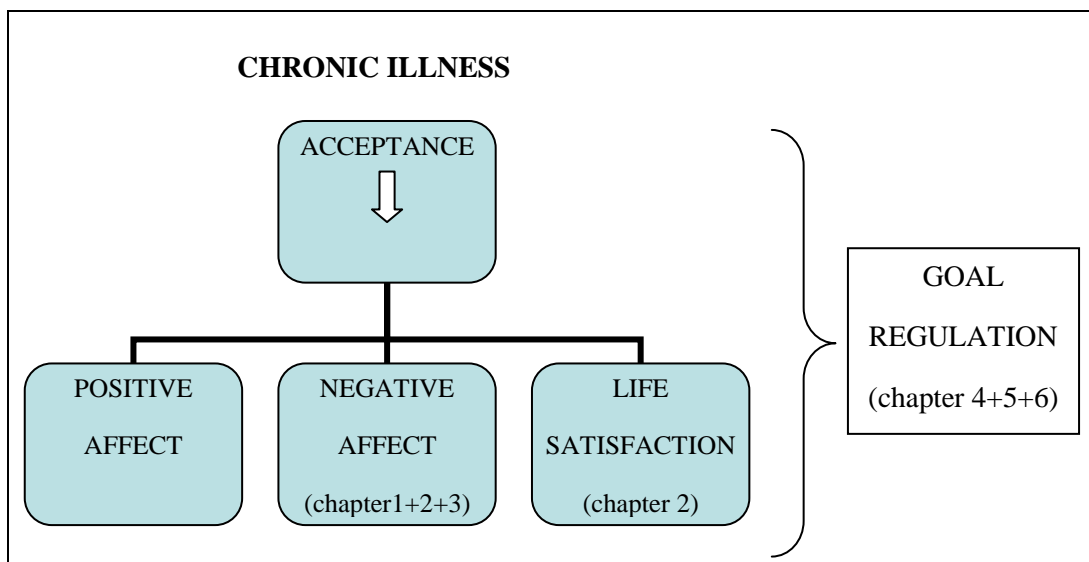


*Figure 1:* Schematic overview dissertation

**REFERENCES**

Aiello, L.P., Gardner, T.W., King, G.L., Blankenship, G., Cavallerano, J.D., Ferris III, F.L., & Klein, R. (1998). Diabetic retinopathy. *Diabetes Care, 21,* 143–156.

Austin, J.T. & Vancouver, J.B. (1996). Goal constructs in psychology: structure, process, and content. *Psychological Bulletin, 120(3),* 338-375.

Ausubel, D.P. (2002). Theory and Problems of Adolescent Development, Third Edition. Lincoln, NE: Writers Club Press.

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Brandstädter, J. & Renner, G. (1990). Tenacious goal pursuit and flexible goal adjustment: explication and age-related analysis of assimilative and accommodative strategies of coping. *Psychology and Aging, 5(1),* 58-67.

Brownson, R.C., Remington, P.L., & Davis, J.R. (1998). *Chronic disease epidemiology and control* (2nd ed.). Washington, DC: American Public Health Association.

Cantor, N., Norem, J., Niedenthal, P.M., Langston, C.A., & Brower, A.M. (1987). Life tasks, self-concept ideals, and cognitive strategies in a life transition. *Journal of Personality and Social Psychology, 53,* 1178-1191.

Carver, C.S. & Scheier, M.F. (1998). On the self-regulation of behaviour. *New York: Cambridge University Press.*

Cross, S., & Markus, H. (1991). Possible selves across the life span. *Human Development*, *34*, 230–255.

Cystic Fibrosis Worldwide (2005). 2005 annual report. Retrieved from http://www.cff.org/aboutCFFoundation/AnnualReport/

Cystic Fibrosis Foundation (2010). Living with cystic fibrosis. Retrieved from http://www.cff.org/LivingWithCF/.

Davies, J.C., Alton, E.W.F.W., & Bush, A. (2007). Cystic fibrosis. *British Medical Journal, 335,* 1255-9. doi: 10.1136/bmj.39391.713229.AD

Davis, P.B. (2006). Cystic fibrosis since 1938. *American Journal of Respiratory and Critical Care Medicine, 173,* 475-482. doi: 10.1164/rccm.200505-840OE

Diener, E. (1984). Subjective well-being. *Psychological Bulletin, 95(3),* 542-575.

Diener, E., Suh, E.M., Lucas, R.E., & Smith, H.L. (1999). Subjective well-being: Three decades of progress. *Psychological Bulletin, 125,* 276-302.

Deci, E. L., & Ryan, R. M. (2000). The "what" and "why" of goal pursuits: Human needs and the selfdetermination of behavior. *Psychological Inquiry, 11,* 227–268.

De Ridder, D. & Kuijer, R. (2007). Reconsidering illness-related goals: is discrepancy resolved by confronting emotions? *Psychology & Health, 22(1),* 107-122. doi: 10.1080/14768320600603471

Diabetes UK (2010). Diabetes in the UK 2010: Key statistics on diabetes. Retrieved from http://www.diabetes.org.uk/Documents/Reports/Diabetes_in_the_UK_2010.pdf

Dijkstra, A., Buunk, A.P., Tóth, G., & Jager, N. (2008). Psychological adjustment to chronic illness: The role of prototype evaluation in acceptance of illness. *Journal of Applied Biobehavioral Research, 12,* 119-140.

Edenborough F.P. (2001). Women with cystic fibrosis and their potential for reproduction. *Thorax 56,* 649–655.

Emmons, R.A. (1986). Personal strivings: An approach to personality and subjective well-being. *Journal of Personality and Social Psychology, 51(5),* 1058-1068.

Emmons, R.A., Colby, P.M., & Kaiser, H.A. (1998). When losses lead to gains: personal goals and the recovery of meaning. In P.T.P. Wong & P.S. Fry (Eds), *The human quest for meaning: a handbook of psychological research and clinical applications* (pp. 163-178). Mahwah, NJ: Lawrence Erlbaum Associates, Inc.

Emmons, R.A. & King, L.A. (1988). Conflict among personal strivings: immediate and long-term implications for psychological and physical well-being. *Journal of Personality and Social Psychology, 54,* 1040-1048.

Erikson, E. H. (1963). Childhood and society (2nd ed.). New York: W.W. Norton & Company.

Evers, A.W.M., Kraaimaat, F.W., van Langenveld, W., Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036.

Ewing, D.J. & Clarke, B.F. (1982). Diagnosis and management of diabetic autonomic neuropathy. *British Medical Journal, 285,* 916-918.

Fagot-Campagna, A., Narayan, K.M., & Imperatore, G. (2001). Type 2 diabetes in children. *British Medical Journal, 322 (7283),* 377-378.

Feinstein, A.B., Forman, E.M., Masuda, A., Cohen, L.L., Herbert, J.D., Moorthy, L.N., & Goldsmith, D.P. (2011). Pain intensity, psychological inflexibility, and acceptance of pain as predictor of functioning in adolescents with juvenile idiopathic arthritis: a

preliminary investigation. *Journal of Clinical Psychology in Medical Settings, 18,* 291-298. doi: 10.1007/s10880-011-9243-6

Frank, R.N. (2004). Diabetic Retinopathy. *The New England Journal of Medicine, 350,* 48-58. Retrieved from www.nejm.org

Gall, M.A., Borch-Johnsen, K., Hougaard, P., Nielsen, F.S., & Parving, H.H. (1995). Albuminuria and poor glycemic control predict mortality in NIDDM. *Diabetes, 44,* 1303–1309.

Gebhardt, W.A. (2006). Contextualising health behaviors: the role of personal goals. In D.T.D. de Ridder & J.B.F. de Wit (Eds.), *Self-regulation in Health Behavior* (pp. 27-43). Chichester (UK): John Wiley & Sons.

Gebhardt, W.A. (2008). On multiple goals and continuous conflict. *The European Health psychologist, 10,* 25-28. Retrieved from www. ehps.net/ehp

Gellman, D. D., Pirani, C. L., Soothill, J. F., Muehrcke, R. C., Maduros, W., & Kark, R. M. (1959). Structure and function in diabetic nephropathy. The importance of diffuse glomerulosclerosis. *Diabetes, 8,* 251-256.

Harris M. & Zimmet P. (1997). Classification of diabetes mellitus and other categories of glucose intolerance. In K. Alberti, P. Zimmet, R. Defronzo (Eds.), *International Textbook of Diabetes Mellitus. Second Edition* (pp. 9-23). Chichester: John Wiley and Sons Ltd.

Henkind, P. (1978). Ocular neovascularization. *American Journal of Ophthalmology, 85,* 287-301.

International Diabetes Federation (2006). *The Diabetes Atlas* (3th ed.). Brussels: International Diabetes Federation.

International Diabetes Federation (2011). Management of diabetes. Retrieved from http://www.idf.org/treatment-diabetes

Kerem, B., Rommens, J.M., Buchanan, J.A., Cox, T.K., Chakravarti, A., Buchwald, M., & Tsui, L.C. (1989). Identification of the cystic fibrosis gene: genetic analysis. *Science, 245 (4922),* 1073-1080. doi: 10.1126/science.2570460

Kuijer, R.G. & De Ridder, D.T.D. (2003). Discrepancy in illness-related goals and quality of life in chronically ill patients: the role of self-efficacy. *Psychology & Health, 18(3),* 313-330. doi: 10.1080/0887044031000146815

Larsson, K., Elding-Larsson, H., Cederwall, E., Kockum, K., Neiderud, J., Sjöblad, S., Lindberg, B., Lernmark, B., Ciolio, C., Ivarsson, S., & Lernmark, A. (2004). Genetic

and perinatal factors as risk factors for childhood type 1 diabetes. *Diabetes/Metabolism Research and Reviews, 20,* 429-437.

Leventhal, H., Brisette, I., & Leventhal, E.A. (2003). The common-sense model of self-regulation of health and illness. In L.D. Cameron & H. Leventhal (Eds.), *The self-regulation of health and illness behaviour* (pp. 42-65). London: Routledge.

Leventhal, H., Halm, E., Horowitz, C., Leventhal, E.A., & Ozakinci, G. (2004). Living with chronic illness: a contextualized, self-regulation approach. In S. Sutton, M. Johnston, & A. Baum (Eds.), *The Sage handbook of health psychology* (pp. 197–240). Thousand Oaks, CA: Sage.

Little, B. R. (1983). Personal projects: A rationale and method for investigation. *Environment and Behavior, 1*5, 273–309.

Little, B.R. (1993). Personal projects and the distributed self: Aspects of a conative psychology. The self in social perspective. In J.M. Suls (Ed.), *The self in social perspective, Psychological perspectives on the self, Vol. 4* (pp. 157-185). Hillsdale, NJ, England: Lawrence Erlbaum Associates.

MacLeod, J.M., Lutale, J., Marshall, S.M. (1995). Albumin excretion and vascular deaths in NIDDM. *Diabetologia, 38,* 610–616.

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: Acceptance and Commitment Therapy for pediatric sickle cell disease. *Journal of Pediatric Psychology, 36,* 398-408. doi:10.1093/jpepsy/jsq118

Maes, S. & Karoly, P. (2005). Self-regulation assessment and intervention in physical health and illness: a review. *Applied Psychology: An International Review, 54(2),* 267-299.

Massey, E.K. (2009). Doctoral Thesis, Leiden University. Retrieved from https://openaccess.leidenuniv.nl/handle/1887/13750

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,* 421-460. doi: 10.1016/j.dr.2008.03.002

Mauer, S.M, Steffes, M.W., Ellis, E.N., Sutherland, D.E.R., Brown, D.M., & Goetz, F.C. (1984). Structural-functional relationships in diabetic nephropathy. *The Journal of Clinical Investigation, 74,* 1143-1155.

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents and young adults with chronic pain: validation of an adapted assessment

instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi:10.1016/j.ejpain.2009.05.002

McGrath, P.J. (1999). Commentary: Headaches in adolescents are a significant problem. *Journal of Pediatric Psychology, 24,* 25-27.

Mogensen, C. E. (1976). Renal functional changes in diabetes. *Diabetes, 25(Suppl. 2),* 872-879.

Mogensen, C.E. (1984). Microalbuminuria predicts clinical proteinuria and early mortality in maturity onset diabetes. *The New England Journal of Medicine, 31,* 356–360.

Moss, S.E., Klein, R., & Klein, B.E. (1988). The incidence of vision loss in a diabetic population. *Ophthalmology, 95,* 1340– 1348.

Nathan, D.M., Buse, J.B., Davidson, M.B., Ferrannini, E., Holman, R.R., Sherwin, R., Zinman, B. (2009). Medical management of hyperglycemia in type 2 diabetes: a consensus algorithm for the initiation and adjustment of therapy. *Diabetes Care, 32(1),* 193-203.

Nathan, D.M., Buse, J.B., Davidson, M.B., Heine, R.J., Holman, R.R., Sherwin, R., & Zinman, B. (2006). Management of hyperglycemia in type 2 diabetes: a consensus algorithm for the initiation and adjustment of therapy. *Diabetes Care, 29(8),* 1963-1972.

Nurmi, J.E. (1987). Age, sex, social class, and quality of family interactions as determinants of adolescents' future orientation: A developmental task interpretation. *Adolescence, 22,* 977–991.

Nurmi, J.E. (1991). How do adolescents see their future? A review of the development of future orientation and planning. *Developmental Review, 11,* 1–59.

Nurmi, J.E. (1992). Age differences in adults' life goals, concerns, and their temporal extension: A life course approach to future-oriented motivation. *International Journal of Behavioral Development*, *15*, 487–508.

Nurmi, J.E. (1993). Adolescent development in an age-graded context: The role of personal beliefs, goals, and strategies in the tackling of developmental tasks and standards. *International Journal of Behavioral Development, 16,* 169–189.

Nurmi, J.E. (2001). Adolescents' self-direction and self-definition in age-graded sociocultural and interpersonal contexts. In J.E. Nurmi (Ed.), *Navigating through adolescence. European perspectives* (pp. 229–249). New York & London: Routledge Falmer.

Nurmi, J.E., Poole, M.E., & Seginer, R. (1995). Tracks and transitions- a comparison of adolescent future-oriented goals, explorations, and commitments in Australia, Israel, and Finland. *International Journal of Psychology, 30(3),* 355-375.

Patrizio, A. & Salameh, W.A. (1998). Expression of the cystic fibrosis transmembrane conductance regulator (CFTR) mRNA in normal and pathological adult human epididymis. *Journal of Reproduction and  Fertility Supplement, l53,* 261-270.

Patterson, C.C., Dahlquist, G.G., Gyürüs, E., Green, A., & Soltész, G. (2009). Incidence trends for childhood type 1 diabetes in Europe during 1989-2003 and predicted new cases 2005-20: a multicentre prospective registration study. *Lancet, 373 (9680),* 2027-2033.

Patz, A. (1982). Clinical and experimental studies on retinal neovascularization. *American Journal of Ophthalmology, 94,* 715-43.

Pinquart, M., & Teubert, D. (2012). Academic, physical, and social functioning o children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 37,* 376-389. doi: 10.1093/jpepsy/jsr106

Pinquart, M., & Shen, Y. (2011). Behavior problems in children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 36,* 1003-1016. doi: 10.1093/jpepsy/jsr042

Quinton, P.M. (1983). Chloride permeability in cystic fibrosis. *Nature, 301 (5899),* 421-422.

Ratjen, F., & Döring, G. (2003). Cystic fibrosis. *The Lancet, 361,* 681-689. Retrieved from www.thelancet.com

Riediger, M. (2001). *On the dynamic relations among multiple goals: Intergoal conflict and intergoal facilitation in younger and older adulthood.* (Doctoral dissertation, Free University, Berlin, Germany). Retrieved from http://www.diss.fu-berlin.de/2001/266/

Riediger, M. & Freund, A.M. (2004). Interference and facilitation among personal goals: differential associations with subjective well-being and persistent goal pursuit. *Personality and Social Psychology Bulletin, 30,* 1511-1523. doi: 10.1177/0146167204271184

Riordan, J. R., Rommens, J. R., Kerem, B. S., Alon, N., Rozmagel, R., Grzelczac, Z., Zielenski, J., Lok, S., Plavsic, N., Chou, J.L., Drumm, M.L., Iannuzzi, M.C., Collins, F.S., & Tsui, L.C. (1989). Identification of the cystic-fibrosis gene − cloning and characterization of complementary-DNA. *Science, 245 (4922),* 1059-1065.

Rommens, J.M., Iannuzzi, M.C., Kerem, B., Drumm, M.L., Melmer, G., Dean, M., Rozmahel, R., Cole, J.L., Kennedy, D., Hidaka, N., Zsiga, M., Buchwald, M., Riordan, J.R., Tsui, L.C., & Collins, F.S. (1989). Identification of the cystic fibrosis gene: chromosome walking and jumping. *Science, 245 (4922),* 1059-1065. Retrieved from www.jstor.org

Rosenstein, B.J., & Zeitlin, P.L. (1998). Cystic fibrosis. *Lancet, 351,* 277-282. Retrieved from www.thelancet.com

Rossing, P., Hougaard, P., Borch-Johnsen, K., & Parving, H.H. (1996). Predictors of mortality in insulin dependent diabetes: 10 year follow-up study. *British Medical Journal, 313,* 779–784

Rothermund, K. (2006). Hanging on and letting go in the pursuit of health goals: Psychological mechanisms to cope with a regulatory dilemma. In de Ridder, D.T.D. & de Wit, J.B.F. (Eds.), *Self-regulation in Health Behavior (*pp. 217-241). Chichester (UK): John Wiley & Sons.

Salmela-Aro, K. & Nurmi, J.E. (1997). Goal contents, well-being, and life context during transition to university: a longitudinal study. *International Journal of Behavioral development, 20(3),* 471-491.

Schwartz, L.A. & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 393-405. doi: 10.1007/s10880-006-9041-8

Schwartz, L.A. & Drotar, D. (2009). Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 34(9),* 954-965. doi:10.1093/jpepsy/jsn144

Seiffge-Krenke, I. (1998). Chronic disease and perceived developmental progression in adolescence. *Developmental Psychology*, *34,* 1073–1084.

The Expert Committee on the Diagnosis and Classification of Diabetes Mellitus (2002). Report of the expert committee on the diagnosis and classification of diabetes mellitus. *Diabetes Care, 25,* S5-S20.

Valmadrid, C.T., Klein, R., Moss, S.E., Klein, B.E. (2000). The risk of cardiovascular disease mortality associated with microalbuminuria and gross proteinuria in persons with older-onset diabetes mellitus. *Archives of Internal Medicine, 160,* 1093–1100. Retrieved from www.archinternmed.com

Viberti, G. C., Pickup, J. C., Jarrett, R. J., & Keen, H. (1979).Effect of control of blood glucose on urinary excretion of albumin and B2-microglobulin in insulin-dependent diabetes. *The New England Journal of Medicine, 300,* 639-641.

Vinik, A.I., Raelene, E.M., Mitchell, B.D., & Freeman, R. (2003). Diabetic autonomic neuropathy. *Diabetes Care, 26,* 1553-1579.

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi:10.1016/j.pain.2011.09.003

Wilson, P.W.F. (1998). Diabetes mellitus and coronary heart disease. *American Journal of Kidney Disease, 32(5),* S89-S100.

World Health Organisation (2006). *10 facts about diabetes*. Retrieved from http://www.who.int/features/factfiles/diabetes/05_en.html

World Health Organisation (2011a). *Diabetes programme*. Retrieved from http://www.who.int/diabetes/en/

World Health Organisation (2011b). *Diabetes*. Retrieved from http://www.who.int/mediacentre/factsheets/fs312/en/index.html

Wrosch, C. & Freund, A.M. (2001). Self-regulation of normative and non-normative developmental challenges. *Human Development, 44,* 264-283.

Yeo, M. & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330,* 721-723. doi: 10.1136/bmj.330.7493.721

Zeltzer, L. (1985). Chronic illness and disability in adolescents. *International Journal of Adolescent Medicine & Health*, *1,* 239–248.

Zhang, X., Saaddine, J.B., Chou, C., Cotch, M.F., Cheng, Y.J., Geiss, L.S., Gregg, E.W., Albright, A.L., Klein, B.E.K., & Klein, R. (2010). Prevalence of diabetic retinopathy in the United States, 2005-2008. *Journal of the American Medical Association, 304(6),* 649-656.

# CHAPTER 1

## THE ROLE OF ACCEPTANCE IN PSYCHOLOGICAL FUNCTIONING IN ADOLESCENTS WITH CYSTIC FIBROSIS: A PRELIMINARY STUDY[1]

**ABSTRACT**

*Introduction:* This study explored the role of acceptance in accounting for the heterogeneity of psychological functioning in adolescents suffering from cystic fibrosis.

*Method:* Thirty-four adolescents completed a battery of questionnaires assessing acceptance, anxiety, depression and disability.

*Results:* Regression analyses revealed that acceptance had a significant and unique contribution in explaining adolescents' anxiety, depression, and disability beyond the effects of demographic variables and parameters of disease severity. Forced expiratory volume, a parameter of disease severity, had a unique contribution in explaining disability, but not in explaining anxiety and depression.

*Conclusions:* Our results support the idea that accepting the limitations imposed by a chronic disease and readjusting life goals, has a positive effect upon psychological functioning in adolescents with cystic fibrosis. Acceptance-based therapies might prove useful in promoting well-being in adolescents with cystic fibrosis.

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**INTRODUCTION**

Cystic fibrosis is a chronic, autosomal-recessive disorder that affects approximately 1 out of 2500 newborn Caucasians (Ratjen & Döring, 2003). Cause of the disease is a mutation in the CFTR-gene, which disturbs the transport of salt and water in and out the body cells (Ratjen & Döring, 2003; Riordan et al., 1989). This results in hyper viscous secretions of the exocrine glands, mainly in the lungs and pancreas (Rosenstein & Zeitlin, 1998). The viscous mucus clogs the airways in the lungs, which often leads to severe lung infection and inflammation (Ratjen & Döring, 2003; Rosenstein & Zeitlin, 1998). The thick mucus also affects the pancreas by blocking the pancreatic ducts, which results in tissue destruction (Ratjen & Döring, 2003). This may lead to an inadequate digestion and malabsorption of nutrients, causing malnutrition, underweight, growth-failure, and, in adolescents, pubertal delay (Ratjen & Döring, 2003).

As yet there is no cure for cystic fibrosis (Cystic Fibrosis Foundation, 2007). Fortunately, effective treatments of the devastating consequences of the mucus exist, and have resulted in a substantial increase in life expectancy (Davis, 2006; Ratjen & Doring, 2003). Despite advances in supportive care, the disease remains associated with physical, psychological and social adversities for the patient and its social environment (Davis, 2006; Gjengedal, Rustøen, Wahl, & Hanestadt, 2003; Wallander & Varni, 1998). Patients still face the possibility of a premature death, and have to undergo an intensive and time-consuming daily treatment (Davis, 2006).

Studies that have investigated the psychological functioning of adolescents with cystic fibrosis reveal variable results. Whereas a number of studies have reported no differences in depressive mood between adolescents with and without cystic fibrosis (Bywater, 1981; Thompson, Hodges, & Hamlet, 1990), other studies have found higher levels of depression (Boyle, di' Sant Agnese, Sack, Millican, & Kulczyckzi, 1976; Pearson, Pumariega, & Seilheimer, 1991). Elevated levels of anxiety, often related to the life-threatening character of the disease (Pfeffer et al., 2003; Willis, Miller, & Wyn, 2001), have been reported in some studies (Boyle et al., 1976; Thompson et al., 1990), but not in others (Pearson et al., 1991; Pfeffer et al., 2003). Mixed results are also found for disability (Badlan, 2006; for a review see Pfeffer et al., 2003). Additional research is needed to clarify these inconsistent findings.

There is clearly a lack of knowledge about the factors that account for the observed heterogeneity in psychological functioning in adolescents with cystic fibrosis. Although studies have investigated the heterogeneity in psychological functioning in

adolescents with chronic physical conditions in general (for a review see Lavigne & Faier-Routman, 1993; Wallander, Varni, 1998), no study has specifically addressed this heterogeneity in adolescents with cystic fibrosis. A potentially important factor in accounting for the heterogeneity in anxiety, depression and disability is *acceptance*. It has been demonstrated that acceptance plays a protective role in the psychological functioning of adults with several chronic diseases such as chronic pain (McCracken, 1998; McCracken & Eccleston, 2003; McCracken & Eccleston, 2005; McCracken, Spertus, Janeck, Sinclair, & Wetzel, 1999; Viane et al., 2003), chronic fatigue syndrome (Van Damme, Crombez, Van Houdenhove, Mariman, & Michielsen, 2006), and multiple sclerosis (Evers, 2001; Pakenham, 2006). Its role has not been explored neither in cystic fibrosis, nor in adolescents. Acceptance can be defined as "recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its aversive consequences" (Evers et al., 2001, p.1027). Accepting cystic fibrosis then means adapting to (1) the demands imposed by the disease, and (2) the limitations the disease involves (Badlan, 2006; Gjengedal et al., 2003). Indeed, cystic fibrosis brings along a number of physical limitations that hinder or even block the realization of certain desired goals. Furthermore, cystic fibrosis requires an intensive, time-consuming daily treatment (Davis, 2006), which may in itself conflict with the pursuit of other valuable goals (Badlan, 2006).

Given the protective potential of acceptance, this study focuses on the role of acceptance in explaining the observed heterogeneity in psychological functioning in adolescents with cystic fibrosis. We focused on the adolescence because this period is crucial in adjusting to disease (Lavigne & Faier-Routman, 1992). In accordance with findings of previous research, we hypothesized that higher levels of acceptance would be related to lower levels of anxiety, depression, and disability. Furthermore, we hypothesized that acceptance would have a unique role in the explaining of psychological functioning, beyond the effects of sociodemographic and disease-related variables.

**METHOD**

**Participants**

Participants were recruited from the University Hospital, Ghent and the CF-Rehabilitation centre, De Haan, Belgium. All adolescents with cystic fibrosis between 12 and 22 years of age who met the following criteria were asked to participate: (1) understanding the Dutch language, and (2) no developmental disorder. Fifty-four

adolescents were invited to participate by letter and were subsequently contacted by phone. A consent was obtained for thirty-four adolescents (response rate = 63%). The main reasons for not participating were a lack of time or motivation and not wanting to be unnecessarily confronted with the disease. Because of confidentiality, no data about the characteristics of the non-participants were available. The study took place in the participants' home, where a written parent consent and/or adolescent assent/consent were obtained, and the questionnaires were filled out. The final sample consisted of eighteen females (52.9%) and sixteen males (47.1%) with cystic fibrosis, all from Caucasian origin. Mean age was 17.31 years (*SD* = 3.05, range 12-22). The majority of the adolescents (82.4%) were attending school. This study was approved by the ethical committee of the University Hospital, Ghent and was carried out in accordance with universal ethical principles (Emanuel, Wendler, & Grady, 2000).

**Measures**

*Acceptance* was assessed by the Illness Cognition Questionnaire (ICQ; Evers et al., 2001). This 18-item questionnaire measures three cognitions about chronic illnesses: 'acceptance', 'helplessness', and 'perceived benefits'. Acceptance is conceptualised as the perceived ability to live with the illness and to master its negative consequences (six items; e.g., "I have learned to live with my illness"). Items are rated on a 4-point Likert scale (1 = 'not at all', 4 = 'completely'). The total score for acceptance varies between 6 and 24, with higher scores indicating higher levels of acceptance. Psychometric research in a sample of adults with rheumatoid arthritis and multiple sclerosis demonstrated the adequate reliability and validity of the ICQ (Evers et al., 2001). Reliability of the acceptance scale in this sample was good with a Cronbach's α of .92.

The Dutch version of the Hospital Anxiety and Depression Scale (HADS; Spinhoven et al., 1997; Zigmond & Snaith, 1983) was used to assess *anxiety and depression.* The HADS consists of 14 items to be rated on a 4-point Likert scale, and has two subscales: 'anxiety' (seven items; e.g., "Do you worry a lot?") and 'depression' (seven items; e.g., "Do you feel optimistic about the future?"). Total scores range between 0 and 21, with higher scores indicating higher levels of anxiety and/or depressive symptoms (Snaith, 2003). For depression, scores between 7 and 9 are indicative of possible depression, scores above 9 of probable depression. Cut-off scores for anxiety are 9 (possible emotional disorder) and 12 (probable emotional disorder) (White, Leach, Sims, Atkinson, & Cottrell, 1999). The HADS is designed for use in medical practice and

has proven reliable and valid as a screening instrument (Spinhoven et al., 1997; Zigmond & Snaith, 1983). Cronbach's α in this study was .70 for anxiety and .80 for depression.

*Disability* was measured with the Dutch version of the Functional Disability Inventory (FDI; Vervoort, Goubert, Eccleston, Bijttebier, & Crombez, 2006; Walker & Greene, 1991), which assesses limitations in physical and psychosocial functioning in children and adolescents with a variety of paediatric conditions (e.g., "doing chores"). The FDI consists of 15 items to be rated on a 5-point Likert scale (0 = 'no difficulty' to 5 = 'impossible'). The total score varies between 0 and 60, higher scores indicating more disability. The reliability and validity of the questionnaire is demonstrated in a sample of children with recurrent and chronic abdominal pain (Claar & Walker, 2006; Walker & Greene, 1991). Cronbach's α for disability in this study was .88.

*Disease severity* was determined by using forced expiratory volume in one second (FEV$_1$% predicted). FEV$_1$% predicted was taken from the medical chart of each adolescent. The most recent pulmonary function test before the administration of the questionnaires was selected. Pulmonary function tests are the standard for measuring respiratory functioning and lung damage for individuals with cystic fibrosis, with lower FEV$_1$% values indicating poorer respiratory functioning and more lung damage (Ramsey & Boat, 1994). Tests were carried out by trained and experienced medical personnel, using a Jaeger MasterScreen Body.

*Sociodemographic information* (age, gender and time since diagnosis) was verbally inquired before administration of the questionnaires.

## RESULTS

### Descriptive statistics and correlations

Means, standard deviations (*SD*'s), and internal consistencies (Cronbach's α) of the questionnaire scores, the time since diagnosis, and FEV$_1$% are presented in Table 1. In the current sample, mean HADS scores were indicative of low levels of psychological distress, with 91.2% of the adolescents scoring beneath the cut-off for anxiety and 88.2% scoring beneath the cut-off for depression (White et al., 1999). For disability, mean scores in the current sample (*M* = 5.42, *SD* = 6.14) were significantly lower in comparison with a sample children with chronic abdominal pain (*M* = 11.25, *t*(628) = 3.42, *p* < .01, two-tailed test) (Claar & Walker, 2006). Mean level of acceptance in the current sample (*M* = 19.06, *SD* = 2.92) was significantly higher than the mean level of acceptance in a sample

of adults with rheumatoid arthritis ($M = 16.65$, $t(295) = 3.24$, $p < .01$, two-tailed test) (Evers et al., 2001).

As expected (see Table 1) there were negative correlations between acceptance and anxiety, depression, and disability. Anxiety was positively associated with depression and disability, as well as depression with disability. $FEV_1\%$ was negatively associated with disability, but not significantly with acceptance, anxiety, and depression. Age, gender, and time since diagnosis were not significantly associated with anxiety, depression, disability, and acceptance.

Table 1

*Means (M), standard deviations (SD), internal consistency (Cronbach's α) and Pearson product-moment correlations of gender, age, time since diagnosis, FEV₁, acceptance, anxiety, depression, and disability*

| Variable | $M$ | $SD$ | α | 2 | 3 | 4 | 5 | 6 | 7 | 8 |
|---|---|---|---|---|---|---|---|---|---|---|
| 1. Gender | - | - | - | .04 | -.03 | -.22 | -.33 | .20 | .09 | .22 |
| 2. Age | 17.31 | 3.05 | - | - | -.15 | -.24 | -.24 | .25 | .16 | .32 |
| 3. Time since diagnosis | 99.28 | 87.19 | - | - | - | .29 | .15 | -.13 | .27 | .15 |
| 4. $FEV_1\%$ | 77.16 | 30.08 | - | - | - | - | .34 | -.22 | -.27 | -.56** |
| 5. Acceptance | 19.06 | 2.92 | .92 | - | - | - | - | -.52** | -.35* | -.47** |
| 6. Anxiety | 4.82 | 2.98 | .70 | - | - | - | - | - | .41* | .58** |
| 7. Depression | 2.35 | 2.99 | .80 | - | - | - | - | - | - | .71** |
| 8. Disability | 5.42 | 6.14 | .88 | - | - | - | - | - | - | - |

*$p < .05$; **$p < .01$

*Note*. $FEV_1$ = forced expiratory volume in one second

*Note*. Time since diagnosis was measured in months

**Regression analyses**

A set of hierarchical regression analyses were performed to examine the unique role of acceptance in explaining anxiety, depression, and disability. In a first step, age and gender were entered into the equation to control for sociodemographic variables. The disease parameters time since diagnosis and $FEV_1\%$ were entered in the second step to control for possible disease-related effects. In the third step, acceptance was entered. Three regression analyses were performed with anxiety, depression, and disability as dependent variables.

The results of the hierarchical regression analyses are reported in Table 2. The demographic variables had no significant contribution in explaining psychological functioning. Of the disease-related parameters, only $FEV_1\%$ had a significant contribution in explaining disability, with an additional 29% of the variance beyond the effects of the

demographic variables. The lower the $FEV_1\%$ value, the higher the degree of reported disability. There was no effect of $FEV_1\%$ upon anxiety and depression. Acceptance made a unique contribution in explaining levels of anxiety, depression, and disability. The additional variance explained by the model incorporating acceptance, was 35% for anxiety, 21% for depression, and 22% for disability. All relations found between acceptance and psychological functioning corroborated the expectations. The higher the degree of acceptance, the lower the degree of anxiety, depression, and disability.

Table 2

*Hierarchical regression analyses of acceptance and anxiety, depression, and disability*

| Dependent variable | Step | Predictor | β | ΔR² | Adj. R² |
|---|---|---|---|---|---|
| Anxiety | 1 | Gender | -.11 | .07 | .01 |
| | | Age | -.03 | | |
| | 2 | Time since diagnosis | -.19 | .04 | -.02 |
| | | $FEV_1\%$ | .05 | | |
| | 3 | Acceptance | -.69** | .35** | .36** |
| Depression | 1 | Gender | -.20 | .01 | -.06 |
| | | Age | -.06 | | |
| | 2 | Time since diagnosis | .27 | .14 | .03 |
| | | $FEV_1\%$ | -.22 | | |
| | 3 | Acceptance | -.54** | .21** | .25** |
| Disability | 1 | Gender | -.12 | .11 | .05 |
| | | Age | .05 | | |
| | 2 | Time since diagnosis | .22 | .29** | .31** |
| | | $FEV_1\%$ | -.45** | | |
| | 3 | Acceptance | -.54** | .22** | .54** |

$*p < .05; **p < .01$

Note: $FEV_1\%$ = forced expiratory volume in one second

Note: Time since diagnosis was measured in months

## DISCUSSION

This study explored the role of acceptance in accounting for psychological functioning in adolescents with cystic fibrosis. We found that acceptance has a unique contribution in explaining anxiety, depression, and disability: adolescents who better accept their disease or, in other words, who feel more able to adapt to the demands (e.g., time-consuming daily treatment) and to the limitations (educational, social and /or professional limitations) imposed by cystic fibrosis, report better psychological functioning. These findings are in line with studies in persons with other chronic health

problems (e.g., chronic pain, chronic fatigue syndrome, multiple sclerosis) or chronic conditions (e.g., deafness, blindness) (Li & Moore, 1998; McCracken, 1998; McCracken & Eccleston, 2003; Pakenham, 2006; Van Damme et al., 2006; Viane et al., 2003). Our findings provide preliminary support for the protective role of acceptance in psychological functioning in cystic fibrosis.

A potential clinical implication of our results is that "learning" to accept the disease may be an important focus in the treatment of adolescents with cystic fibrosis. Acceptance-based interventions already exist for the treatment of several physical and psychological disorders such as depression (Ma & Teasdale, 2004), psychotic symptoms (Bach & Hayes, 2002), cancer (Speca, Carlson, Goodey, & Angen, 2000), and chronic pain (Kabat-Zinn, Lipworth, & Burney, 1985). Research into the effectiveness of these interventions reveals promising and positive results (McCracken & Eccleston, 2003).

A number of limitations of the study deserve consideration. First, this is the first study addressing acceptance in adolescents with cystic fibrosis, and further research is needed to corroborate our findings and to establish whether our results generalize to other samples. Second, our sample of adolescents with cystic fibrosis reported relatively few psychological difficulties. More research is warranted to investigate the potential reasons for these low levels of disability and psychological difficulty. Possible reasons can be sample bias or psychological mechanisms such as denial, defensiveness and repressive coping. Third, only self-report measures administered to the adolescents were used. In future studies, assessment from additional informants, such as parents, may be incorporated to obtain extra information on the psychological functioning of these adolescents. Fourth, because of a lack of instruments assessing acceptance and a lack of instruments specially developed for assessing anxiety and depression for medical problems, acceptance, anxiety and depression were only assessed using single scales. Fifth, the sample size of the present study was small. Possibly only the strong effects sizes were observed, while smaller effects remained undetected. Sixth, findings were based on cross-sectional and correlational data, and hence, do not indicate causal effects. Although the current results might indicate that acceptance leads to a better psychological functioning, also the reverse might be true. It seems reasonable that individuals who experience minor anxiety, depression, and disability will have less difficulty adapting to the negative consequences of cystic fibrosis and therefore will exhibit higher acceptance. Research in samples with worse functioning is required to clarify this issue. Also longitudinal research is necessary to get a more detailed picture of the interplay between

acceptance and psychological functioning. Seventh, participants included in this study have a broad age range, by which possible developmental differences in acceptance and/or psychological functioning can be concealed. Further research with more homogenous groups is needed to examine this issue and to confirm the current findings among different age groups.

Despite these limitations the results of this study provide preliminary support for the role of acceptance in adolescents with cystic fibrosis. As this is, to our knowledge, the first study investigating this role in cystic fibrosis, additional research is needed to confirm and elaborate the current findings.

REFERENCES

Bach, P., & Hayes, S.C. (2002). The use of acceptance and commitment therapy to prevent the rehospitalisation of psychotic patients: a randomized controlled trial. *Journal of Consulting and Clinical Psychology, 70(5),* 1129-1139. doi: 10.1037//0022-006X.70.5.1129

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Boyle, I.R., di Sant'Agnese, P.A., Sack, S., Millican, F., & Kulczycki, L.L. (1976). Emotional adjustment of adolescents and young adults with cystic fibrosis. *The Journal of Pediatrics, 88,* 318-326.

Bywater, E.M. (1981). Adolescents with cystic fibrosis: Psychosocial adjustment. *Archives of Disease in Childhood, 56,* 538-543.

Claar, R.L., & Walker, L.S. (2006). Functional assessment of pediatric pain patients: Psychometric properties of the Functional Disability Inventory. *Pain, 121,* 77-84. doi: 10.1016/j.pain.2005.12.002

Cystic Fibrosis Foundation (2007). About Cystic Fibrosis. Retrieved from http://www.cff.org/AboutCF/

Damme, S. Van, Crombez, G., Houdenhove, B. Van, Mariman, A., & Michielsen, W. (2006). Well-being in patients with chronic fatigue syndrome: The role of

acceptance. *Journal of Psychosomatic Research, 61,* 595-599. doi:10.1016/j.jpsychores.2006.04.015

Davis, P.B. (2006). Cystic fibrosis since 1938. *American Journal of Respiratory and Critical Care Medicine, 173,* 475-482. doi: 10.1164/rccm.200505-840OE

Emanuel, E.J., Wendler, D., & Grady, C. (2000). What makes clinical research ethical? *Journal of the American Medical Association, 283,* 2701-2711. Retrieved from www.jama.com

Evers, A.W.M., Kraaimaat, F.W., Lankveld, W. van, Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavorable thinking: The Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Gjengedal, E., Rustøen, T., Wahl, A.K., & Hanestadt, B.R. (2003). Growing up and living with cystic fibrosis. Everyday life and encounters with the health care and social services – A qualitative study. *Advances in Nursing Science, 26 (2)*, 149-159.

Kabat-Zinn, J., Lipworth, L., & Burney, R. (1985). The clinical use of mindfulness meditation for the self-regulation of chronic pain. *Journal of Behavioral Medicine, 8*, 163-190.

Lavigne, J.V. & Faier-Routman, J. (1992). Psychological adjustment to pediatric physical disorders: A meta-analytic review. *Journal of Pediatric Psychology, 17,* 133-157. doi: 10.1093/jpepsy/17.2.133

Lavigne, J.V. & Faier-Routman, J. (1993). Correlates of psychological adjustment to pediatric physical disorders: A meta-analytic review and comparison with existing models. *Developmental and Behavioral Pediatrics, 14,* 117-123.

Li, L., & Moore, D. (1998). Acceptance of disability and its correlates. *The Journal of Social Psychology, 138,* 13-25.

Ma, H.S., & Teasdale, J.D. (2004). Mindfulness-based cognitive therapy for depression: replication and exploration of differential relapse prevention effects. *Journal of Consulting and Clinical Psychology, 72(1),* 31-40. doi: 10.1037/0022-006X.72.1.31

McCracken, L.M. (1998). Learning to live with the pain: Acceptance of pain predicts adjustment in persons with chronic pain. *Pain, 74,* 21-27. doi: 10.1016/S0304-3959(97)00146-2

McCracken, L.M., & Eccleston, C. (2003). Coping or acceptance: What to do about chronic pain? *Pain, 105,* 197-204. doi: 10.1016/S0304-3959(03)00202-1

McCracken, L.M., & Eccleston, C. (2005). A prospective study of pain and patient functioning with chronic pain. *Pain, 118,* 164-169. doi: 10.1016/j.pain.2005.08.015

McCracken, L.M., Spertus, I.L., Janeck, A.S., Sinclair, D., & Wetzel F.T. (1999). Behavioral dimensions of adjustment in persons with chronic pain: Pain-related anxiety and acceptance. *Pain, 80,* 283-289. doi: 10.1016/S0304-3959(98)00219-X

Pakenham, K.I. (2006). Investigation of the coping antecedents to positive outcomes and distress in multiple sclerosis (MS). *Psychology and Health, 21(5),* 633-649. doi: 10.1080/14768320500422618

Pearson, D.A., Pumariega, A.J., & Seilheimer, D.K. (1991). The development of psychiatric symptomatology in patients with cystic fibrosis. *Journal of the American Academy of Child and Adolescent Psychiatry, 30,* 290-297. doi: 10.1097/00004583-199103000-00019

Pfeffer, P.E., Pfeffer, J.M., & Hodson, M.E. (2003). The psychological and psychiatric side of cystic fibrosis in adolescents and adults. *Journal of Cystic Fibrosis, 2,* 61-68. doi:10.1016/S1569-1993(03)00020-1

Ramsey, B.W., & Boat, T.F. (1994). Outcome measures for clinical trials in cystic fibrosis. *The Journal of Pediatrics, 124,* 177-192. doi: 10.1016/S0022-3476(94)70301-9

Ratjen, F., & Döring, G. (2003). Cystic fibrosis. *The Lancet, 361,* 681-689. doi: 10.1016/S0140-6736(03)12567-6

Riordan, J.R., Rommens, J.R., Kerem, B.S., Alon, N., Rozmagel, R., Grzelczac, Z., Zielenski, J., Lok., S., Plavsic, N., Chou, J.L., Drumm, M.L., Iannuzzi, M.C., Collins, F.S., & Tsui, L.C. (1989). Identification of the cystic-fibrosis gene – Cloning and characterization of complementary-dna. *Science, 245 (4922),* 1066-1072.

Rosenstein, B.J., & Zeitlin, P.L. (1998). Cystic fibrosis. *Lancet, 351,* 277-282. doi: 10.1016/S0140-6736(97)09174-5

Snaith, P.R. (2003). The Hospital Anxiety and Depression Scale. *Health and Quality of Life Outcomes, 1,* 29-32.

Speca, M., Carlson, L.E., Goodey, E., & Angen, M. (2000). A randomized, wait-list controlled clinical trial: the effect of a mindfulness meditation-based stress reduction program on mood and symptoms of stress in cancer outpatients. *Psychosomatic Medicine, 62,* 613-622.

Spinhoven, P., Ormel, J., Sloekers, P.P.A., Kempen, G.I.J.M., Speckens, A.E.M., & Hemert, A.M. Van (1997). A validation study of the Hospital Anxiety and Depression Scale (HADS) in different groups of Dutch subjects. *Psychological Medicine, 27,* 363-370. Retrieved from http://journals.cambridge.org

Thompson, R.J., Hodges, K., & Hamlet, K.W. (1990). A matched comparison of adjustment in children with cystic fibrosis and psychiatrically referred and nonreferred children. *Journal of Pediatric Psychology, 15,* 745-759. doi: 10.1093/jpepsy/15.6.745

Vervoort, T., Goubert, G., Eccleston, C., Bijttebier, P., & Crombez, G. (2006). Catastrophic thinking about pain is independently associated with pain severity, disability, and somatic complaints in school children and children with chronic pain. *Journal of Pediatric Psychology, 31(7),* 674-683. doi: 10.1093/jpepsy/jsj059

Viane, I., Crombez, G., Eccleston, C., Poppe, C., Devulder, J., Houdenhove, B. Van, & Corte, W. De (2003). Acceptance of pain is an independent predictor of mental well-being in patients with chronic pain: Empirical evidence and reappraisal. *Pain, 106,* 65-72. doi:10.1016/S0304-3959(03)00291-4

Walker, L.S., & Greene, J.W. (1991). The Functional Disability Inventory: Measuring a neglected dimension of child health status. *Journal of Pediatric Psychology, 16,* 39-58. doi: 10.1093/jpepsy/16.1.39

Wallander, J.L., & Varni, J.W. (1998). Effect of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry, 39,* 29-46. doi: 10.1017/S0021963097001741

White, D., Leach, C., Atkinson, M., & Cottrell, D. (1999). Validation of the Hospital Anxiety and Depression Inventory for use with adolescents. *British Journal of Psychiatry, 175,* 452-454.

Willis, E., Miller, R., & Wyn, J. (2001). Gendered embodiment and survival of young people with cystic fibrosis. *Social Science and Medicine, 53,* 1163-1174. doi: 10.1016/S0277-9536(00)00416-0

Zigmond, A.S., & Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavia, 67,* 361-370.

# CHAPTER 2

## ACCEPTANCE AND WELL-BEING IN ADOLESCENTS AND YOUNG ADULTS WITH CYSTIC FIBROSIS: A PROSPECTIVE STUDY[1]

**ABSTRACT**

*Objective:* To prospectively investigate the role of acceptance in well-being in adolescents and young adults with cystic fibrosis.

*Method:* Forty adolescents and young adults with cystic fibrosis (ages 14-22) completed questionnaires assessing acceptance, anxiety and depressive symptoms, physical functioning, role functioning, emotional functioning, and social functioning. After six months, 28 of them completed the questionnaires on anxiety and depressive symptoms, physical functioning, role functioning, emotional functioning, and social functioning a second time.

*Results:* More acceptance (Time 1) was related to less depressive symptoms (Time 1 and 2), and to better role, emotional, and social functioning (Time 1).

*Conclusions:* Results indicate that accepting the limitations imposed by chronic disease and readjusting life goals may have a positive effect upon well-being in adolescents and young adults with cystic fibrosis. Further research is needed to clarify whether acceptance-based interventions are useful in promoting well-being in adolescents and young adults with cystic fibrosis.

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**INTRODUCTION**

Cystic fibrosis (CF) is the most common life-shortening genetic disease amongst the Caucasian population, affecting 1 in 2500 newborn children (Cystic Fibrosis Worldwide, 2005; Ratjen & Döring, 2003). The disease is marked by an increased viscosity of the exocrine secretions throughout the body. These hyper viscous secretions mainly affect the lungs and the pancreas by plugging the airways and pancreatic ducts. Mucus plugging then triggers recurrent cycles of infection and inflammation what results in irreversible tissue damage (Ratjen & Döring, 2003; Rosenstein & Zeitlin, 1998). Given the physical consequences of the disease, CF also has negative consequences on other domains of functioning (Glasscoe & Quittner, 2008).

Due to substantial improvements in specialised care, almost half of the children and adolescents with CF nowadays survive into adulthood (Cystic Fibrosis Foundation, 2010). As a consequence, attention to their long-term well-being has become increasingly important (Badlan, 2006; Glasscoe & Quittner, 2008). Well-being refers to a broad area of experiences that encompasses positive and negative affect, general life satisfaction, and satisfaction with specific domains of life (Diener, 1984; Diener, Suh, Lucas, & Smith, 1999). With regard to well-being, adolescents and young adults with CF seem to be confronted with two major challenges: managing the affective impact of CF and maintaining a satisfactory life (Glasscoe & Quittner, 2008; Quittner et al., 2008; Schwartz & Drotar, 2009). Crucial in handling these challenges is to balance the limitations (e.g., functional disability) and demands of having CF (e.g., disease management) with the pursuit of important developmental goals in order to pass on to adulthood (e.g, planning for the future, becoming independent, forming close relationships, preparing for a job, making decisions about family life, becoming socially responsible, and moving away from home) (Badlan, 2006; Gjengedal et al., 2003; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009). Examples of this balance may be the adjustment of developmental goals that have become unrealistic due to CF (e.g., working part-time instead of full-time, postpone university studies and first get health back on track, only going out in non-smoking places) or the integration of disease management and important developmental goals (e.g., doing aerosol therapy while studying). For individuals with CF, adolescence is a particularly challenging period as from then on the limitations and demands of having CF typically increase (Ernst et al., 2010). Therefore, it is essential to understand how adolescents and young adults with CF maintain their well-being despite the growing burden of having CF.

Despite the finding that the challenges of having CF can severely impact upon well-being (Glasscoe & Quittner, 2008; Quittner et al., 2008; Riekert, Bartlett, Boyle, Krishnan, & Rand, 2007; Schwartz & Drotar, 2009), research has shown that not all individuals with CF struggle to maintain well-being. Indeed, while some research findings indicate that adolescents and young adults with CF experience difficulties in several components of well-being such as elevated levels of depression (Quittner et al., 2008), elevated levels of anxiety (Pfeffer, Pfeffer, & Hodson, 2003), and an affected quality of life (Goldbeck & Schmitz, 2001; Pfeffer et al., 2003), other findings indicate no significant differences between adolescents and young adults with and without CF (Anderson, Flume, & Hardy, 2001; Bregnballe, Thastum, & Schiotz, 2007; Goldbeck & Schmitz, 2001; Havermans, Colpaert, & Dupont, 2008; Pfeffer et al., 2003; Szyndler, Towns, van Asperen, & McKay, 2005). Hence, understanding why certain adolescents and young adults maintain their well-being despite the burden of CF is of utmost importance.

A process that may account for the individual variability in well-being is *acceptance*. Acceptance can be defined as "recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences" (Evers et al., 2001, p.1027). Acceptance thus comprises the ability to reconcile to the limitations the disease/illness involves, and to face the variable demands imposed by the disease, while staying engaged in a valuable life (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003). As CF is an incurable disease with concomitant limitations, acceptance may be a key factor in the well-being of affected individuals (Ernst, Johnson, & Stark, 2010).

The role of acceptance in well-being has already been examined in several chronic conditions. Among adults with chronic pain (e.g., McCracken, Carson, Eccleston, & Keefe, 2004; McCracken & Vowles, 2008; McCracken & Zhao-O'Brien, 2010; Viane et al., 2003), cancer pain (Gauthier et al., 2009), chronic fatigue syndrome (Van Damme, Crombez, Van Houdenhove, Mariman, & Michielsen, 2006), tinnitus (Westin, Hayes, & Andersson, 2008), and multiple sclerosis (Evers et al., 2001; Pakenham, 2006), acceptance was significantly associated with positive outcomes such as less anxiety and depression, better mood, better emotional, social, and physical functioning, less physical complaints, better work status, and less disability. To our knowledge, only one study examined acceptance in adolescents and young adults with CF, demonstrating that acceptance was related to less anxiety, depression and disability (Casier et al., 2008). This

study, however, was cross-sectional and did not focus on well-being in general. The present study extends this research by investigating the long-term effects of acceptance on well-being. Identifying the effects of acceptance on well-being over time is important as it may point to processes that promote a valuable and meaningful life in the context of a life-shortening illness (Ernst et al., 2010). Furthermore, investigating the associations between acceptance and well-being may provide indications whether acceptance-based interventions (Hayes, 2004; Hayes et al., 2006) are useful for the preservation and enhancement of these adolescents' and young adults' well-being, and, if so, these findings may further guide the development of these interventions in the context of CF (Hayes, 2004; Hayes et al., 2006).

Well-being was operationalised in terms of anxiety and depressive symptoms, and health-related quality of life. These are important components of well-being, and can be detrimentally affected in individuals with CF (Anderson et al., 2001; Diener et al., 1999; Pfeffer et al., 2003; Quittner et al., 2008; White, Miller, Smith, & McMahon, 2009). In line with expert consensus, health-related quality of life was operationalised as a subjective construct that fundamentally includes four core domains: physical functioning, emotional functioning, social functioning, and role functioning (Cella, 1998; Spilker, 1996). We hypothesized that higher levels of adolescents' and young adults' acceptance at Time 1 are related to less anxiety and depressive symptoms, and to better physical, emotional, social, and role functioning at both Time 1 and 2 (6 months later) (see Evers et al., 2001; McCracken, et al., 2004; Pakenham, 2006).

## METHOD

### Participants

All adolescents and young adults with CF attending the University Hospitals of Antwerp, Brussels, or Ghent, and the Sint-Vincentius Hospital, Antwerp, Belgium who were 14-22 years of age and met the following criteria were invited to participate: understanding the Dutch language, no developmental disorder, and no (planned) lung transplantation. Adolescents and young adults who had/were awaiting lung transplantation were not included as they find themselves in a situation that is very different from the situation of individuals in the pre-transplant period (e.g., very high level of symptoms, being terminally ill, intertwinement of end of life issues and hope of transplantation, risk of post-transplant complications, dramatic change of illness status) (Bourke et al., 2009; Kurland & Orenstein, 2001). Seventy-three adolescents and young

adults met these criteria. The participating centres did not contact nine of these adolescents and young adults because they had serious physical and/or psychosocial problems. Sixty-four adolescents and young adults were invited to participate by letter and/or approached during routine clinic visits by the psychologist from the respective hospital (time period of recruitment: 13 months). Subsequently, adolescents and young adults agreeing to participate were contacted by phone by a research assistant. Forty adolescents and young adults (23 boys; 17 girls) were enrolled in our study (response rate = 62.50 %). The main reasons for not participating were a lack of time or motivation ($N =$ 14), too many requests for participating in scientific research ($N = 7$), and not wanting to be unnecessarily confronted with the disease ($N = 2$). Mean age was 18.40 years ($SD =$ 2.87). To determine disease severity, the US CF Foundation guidelines for severity of lung disease were used (Cystic Fibrosis Foundation, 2008). Disease severity ranged from not severe/normal lung function (FEV$_1$% predicted $\geq$ 90%) to severe (FEV$_1$% predicted < 40%). Mean disease severity indicated mild lung disease ($M_{FEV1\%}$ = 83.01%). Of all adolescents and young adults, 32.50% had normal lung function (FEV$_1$% predicted $\geq$ 90%), 42.50% mild lung disease (89% $\geq$ FEV$_1$% predicted $\geq$ 70%), 22.50% moderate lung disease (69% $\geq$ FEV$_1$% predicted $\geq$ 40%), and 2.5% had severe lung disease (FEV$_1$% predicted < 40%). Mean time since diagnosis was 16.84 years ($SD = 3.82$). All adolescents and young adults were of Caucasian origin. Because of confidentiality, no data about the characteristics of the non-participants were available.

**Procedure**

At Time 1, a research assistant visited the participants at their home. During this house visit, written parent consent and/or adolescents and young adult assent/consent were obtained, and a booklet of questionnaires (assessing acceptance, anxiety, depression, and health-related quality of life) were filled out. At Time 2, all adolescents and young adults were contacted again and asked to complete follow-up questionnaires (assessing anxiety, depression, and health-related quality of life). The follow-up questionnaires were sent by mail. Twenty-eight of the 40 adolescents and young adults (70%) returned the Time 2 questionnaires. Drop-out analyses showed that there were no significant differences in age ($t(38) = -1.03$, *ns*), disease severity ($t(38) = -.61$, *ns*), acceptance ($t(38)$ = -1.48, *ns*), anxiety ($t_{anxiety}(38) = .64$, *ns*), depression ($t_{depression}(38) = 1.13$, *ns*), and health-related quality of life ($t_{physical\ functioning}(38) = -.98$, *ns*; $t_{role\ functioning}(38) = -1.08$, *ns*; $t_{emotional\ functioning}(38) = -1.91$, *ns*; $t_{social\ functioning}(38) = -.92$, *ns*) between the adolescents and young adults who responded at Time 2 and those who did not. This study was approved by the

ethical committees of the University Hospitals of Antwerp, Brussels, and Ghent, and the Sint-Vincentius Hospital, Antwerp and was carried out in accordance with universal ethical principles (Emanuel, Wendler, & Grady, 2000).

**Measures**

*Acceptance* was assessed by the Dutch version of the Illness Cognition Questionnaire (ICQ; Evers et al., 2001). This 18-item questionnaire contains three subscales: 'acceptance', 'helplessness', and 'perceived benefits'. Only the acceptance scale was used. Acceptance is conceptualised as the perceived ability to live with the illness and to master its negative consequences (six items; e.g., "I can handle the problems related to my illness"). Items are rated on a 4-point Likert scale (1 = 'not at all', 4 = 'completely'). The total score for acceptance varies between 6 and 24, with higher scores indicating higher levels of acceptance. Psychometric research in samples of Dutch-speaking adults with rheumatoid arthritis, multiple sclerosis, chronic pain and chronic fatigue demonstrated the adequate reliability and validity of the ICQ (Evers et al., 2001; Lauwerier et al., 2010). Reliability of the acceptance scale in this sample was good with a Cronbach's α of .90 (see Table 1).

*Health-related quality of life* was assessed by the Dutch version of the Cystic Fibrosis Questionnaire-Revised Teen/Adult Version (CFQ-R Teen/Adult Version; Klijn et al., 2004; Quittner, Modi, Watrous, & Davis, 2003). The CFQ-R Teen/Adult Version consists of 50 items assessing nine quality of life domains and is sensitive to the specific concerns of individuals (aged 14 and older) with CF: 'physical functioning', 'role functioning', 'vitality', 'emotional functioning', 'social functioning', 'body image', 'eating', 'treatment burden', and 'overall health perception'. The CFQ-R Teen/Adult Version also assesses three symptom scales: 'weight', 'respiratory', and 'digestion'. Because no higher order summary scores are available, only the subscales measuring the core domains of health-related quality of life were included. In line with expert consensus (Cella, 1998; Spilker, 1996), the following subscales were used: physical functioning (eight items; e.g., "Difficulty to walk as fast as others"), role functioning (four items, e.g., "Trouble keeping with school, work or daily activities"), emotional functioning (five items; e.g., "Felt worried"), and social functioning (six items; e.g. "Stayed at home more than you wanted) (Quittner  et al., 2003; Quittner, Buu, Messer, Modi, & Watrous, 2005). Items are rated in terms of a frequency response on a 4-point Likert scale (1 = 'all the time', 4 = 'never'), a difficulty rating on a 4-point Likert scale (1 = 'a lot of difficulty', 4 = 'no difficulty'), a true-false response on a 4-point Likert scale (1 = 'very true', 4 =

'very false'), or a selection of a particular statement (4- or 5-point Likert scale). Total scores vary from 0 to 100, with higher scores representing better quality of life (Quittner et al., 2000). Reported psychometric properties of the CFQ-R Teen/Adult Version subscales were average to good (Quittner et al., 2003). In the current sample, the internal consistencies (Cronbach's α) at Time 1 were .92 for physical functioning, .81 for role functioning, .63 for emotional functioning, and .44 for social functioning. Internal consistencies (Cronbach's α) at Time 2 were .84 for physical functioning, .37 for role functioning, .55 for emotional functioning, and .60 for social functioning (see Table 1).

The Dutch version of the Hospital Anxiety and Depression Scale (HADS; Spinhoven et al., 1997; Zigmond & Snaith, 1983) was used to assess *anxiety and depressive symptoms*. The HADS consists of 14 items to be rated on a 4-point Likert scale, and has two subscales: 'anxiety' (seven items; e.g., "Do you feel tense and wound up?") and 'depression' (seven items; e.g., "Do you feel cheerful?"). Total scores range between 0 and 21, with higher scores indicating higher levels of anxiety and/or depressive symptoms (Snaith, 2003). For depression, scores between 7 and 9 are indicative of possible depression, scores above 9 of probable depression. Cut-off scores for anxiety are 9 (possible emotional disorder) and 12 (probable emotional disorder) (White, Leach, Sims, Atkinson, & Cottrell, 1999). The HADS is designed for use in medical practice. It is proven reliable and valid as a screening instrument in adolescents, adults, and elderly subjects with or without a medical condition (Spinhoven et al., 1997; White et al., 1999; Zigmond & Snaith, 1983). Cronbach's α in this study for anxiety was .71 at Time 1 and 2. For depression Cronbach's α  was .73 at Time 1 and .76 at Time 2 (see Table 1).

*Disease severity* was determined by using forced expiratory volume in one second (FEV$_1$% predicted). FEV$_1$% predicted was taken from the medical chart of each adolescent or young adult. The most recent pulmonary function test before the administration of the questionnaires was selected ($M_{days}$ = 35.56, *SD* = 27.29, range 1 – 106 days). Pulmonary function tests are the standard for measuring respiratory functioning and lung damage for individuals with cystic fibrosis. FEV$_1$% predicted, was used as a continuous variable, with lower values indicating poorer respiratory functioning and more lung disease (Ramsey & Boat, 1994). Tests were carried out by trained and experienced medical personnel, using a Jaeger MasterScreen$^{TM}$ Body (CareFusion Germany 234 GmbH, Hoechberg, Germany). During this procedure, individuals are seated in a glass cabin where they perform inspiratory and expiratory manoeuvres through a mouthpiece (CareFusion, 2009).

*Socio-demographic information* (age, gender and time since diagnosis) was verbally inquired before administration of the questionnaires.

**Statistical analyses**

All analyses were performed using SPSS 15.0. Tests of normality were performed for all continuous variables. Descriptive statistics were computed for all variables of interest. Pearson's correlations were performed to determine whether age, time since diagnosis, disease severity and acceptance were related to anxiety and depressive symptoms, and health-related quality of life. Furthermore, *t*-tests were performed to examine gender differences in anxiety and depressive symptoms, and/or health-related quality of life, and to identify potential differences between the Time 1 and Time 2 dependent variables.

Separate hierarchical regression analyses were used to investigate the role of acceptance (Time 1) in anxiety and depressive symptoms (Time 1 and 2) and health-related quality of life (Time 1 and 2). For regressions with the dependent variable measured at Time 1, socio-demographic and disease-related variables that were significantly related to the outcome measures were entered as control variables in a first step. Acceptance was entered in the second step. Anxiety and depressive symptoms, and physical, role, emotional, and social functioning at Time 1 were entered as dependent variables. For regressions with the dependent variable measured at Time 2, socio-demographic and disease-related variables that were significantly related to the outcome measures were entered as control variables in a first step. In the second step, we controlled for the corresponding Time 1 measure of respectively anxiety and depressive symptoms, and physical, role, emotional, and social functioning. In the third step, acceptance was entered. Anxiety and depressive symptoms, and physical, role, emotional, and social functioning measured at Time 2 were used as dependent variables. As we had a priori hypotheses about the direction of effects, one-tailed tests of significance were used (Kline, 2004; Martin & Bateson, 1993, 2007; Wonnacott & Wonnacott, 1985). Alpha level was set at $p < .05$. To control for multiple testing and balance the amount of type I and type II errors, the Benjamini and Hochberg false discovery rate (i.e., the expected proportion of rejected true null hypothesis among rejected hypotheses) was used (Benjamini & Hochberg, 1995; Benjamini, Krieger, & Yekutieli, 2006). The False Discovery Rate level was set at 5%, assuring that in each set of hierarchical regression analyses (i.e., analyses at Time 1 and at Time 2) no more than 5% of the significant relationships found are false-positive (Benjamini et al., 2006).

The initial sample ($N = 40$) had sufficient power (.86) to detect large effects (see Casier et al., 2008) in multiple regressions ($f^2 = .35$) with three variables. The second sample ($N = 28$) had a power of .65 to detect large effects in multiple regressions ($f^2 = .35$) with three variables (Vacha-Haase & Thompson, 2004).

## RESULTS

### Descriptive statistics

Means, standard deviations, and internal consistencies (Cronbach's α) of variables are reported in Table 1. The mean score for acceptance was 18.55 ($SD = 4.16$), which is in line with a previous study on acceptance in adolescents and young adults with CF (Casier et al., 2008). Mean scores for anxiety and depressive symptoms were indicative of low psychological distress. At Time 1, 85 % of the adolescents and young adults scored beneath the cut-off for probable anxiety, and 97.50 % beneath the cut-off for probable depression (White et al., 1999). The percentage of adolescents and young adults scoring beneath this cut-off at Time 2 was 88.90 % for anxiety and 96.30 % for depression. Mean levels of physical, role, emotional, and social functioning varied between 71.39 and 85.71. Paired sample *t*-tests revealed that there were no significant differences between the Time 1 and Time 2 dependent variables. Tests of normality revealed that none of the variables included were skewed.

### Identification of relevant control variables

Correlations between age, time since diagnose, disease severity, acceptance and anxiety and depressive symptoms, and health-related quality of life are reported in Table 1. Socio-demographic and disease-related variables that were significantly related to the outcome measures were entered as control variables in the hierarchical regression analyses. Furthermore, t-tests revealed that gender was related to emotional functioning at Time 1 ($M_{male} = 78.84$, $SD_{male} = 13.43$; $M_{female} = 67.84$, $SD_{female} = 19.18$; $F(1,38) = 4.56$, $p = .04$), and to social functioning at Time 1 ($M_{male} = 75.85$, $SD_{male} = 11.68$; $M_{female} = 65.36$, $SD_{female} = 17.07$; $F(1,38) = 5.33$, $p = .03$).

Table 1

*Means (M), standard deviations (SD), internal consistency (Cronbach's α) and Pearson product-moment correlations of acceptance (Time 1), anxiety, depression, physical functioning, role functioning, emotional functioning, and social functioning. Correlation coefficients above the diagonal are at Time 1, coefficients beneath are with the outcomes measured at Time 2*

| | Time 1 | | Time 2 | | | | | | | | | | | |
| | M(SD) | α | M(SD) | α | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1. Age | 18.40 (2.87) | - | 18.70 (2.80) | - | - | .73* | -.45* | -.03 | .02 | .10 | -.06 | -.22 | -.21 | .05 |
| 2. Time since diagnosis | 16.84 (3.82) | - | 16.99 (4.07) | - | .69* | - | -.43* | -.07 | -.03 | .17 | -.08 | -.16 | -.28* | -.02 |
| 3. Disease severity | 83.01 (19.7) | - | 84.26 (16.34) | - | -.31 | -.36* | - | -.10 | -.14 | -.21 | .32* | .34* | .14 | .07 |
| 4. Acceptance | 18.55 (4.16) | .90 | - | - | -.14 | -.14 | -.16 | - | -.29* | -.35* | .15 | .29* | .45* | .54* |
| 5. Anxiety | 4.88 (3.49) | .71 | 3.79 (4.04) | .71 | .18 | .06 | -.27 | -.12 | - | .49* | -.49* | -.56* | -.61* | -.44* |
| 6. Depression | 1.53 (2.05) | .73 | 1.07 (2.96) | .76 | .12 | -.03 | -.03 | -.35* | .65* | - | -.52* | -.58* | -.57* | -.53* |
| 7. Physical functioning | 81.46 (20.04) | .92 | 85.71 (13.82) | .84 | -.18 | -.23 | .54* | .05 | -.41* | -.25 | - | .50* | .59* | .39* |
| 8. Role functioning | 83.13 (17.95) | .81 | 85.42 (11.02) | .37 | -.28 | -.05 | .36* | -.08 | -.16 | -.14 | .36* | - | .55* | .32* |
| 9.Emotional functioning | 74.17 (16.82) | .63 | 76.19 (12.63) | .55 | -.10 | -.01 | .22 | .05 | -.51* | -.22 | .50* | .24 | - | .68* |
| 10. Social functioning | 71.39 (14.97) | .44 | 74.01 (13.70) | .59 | .00 | .11 | .22 | .34* | -.42* | -.65* | .38* | .22 | .43* | - |

*One-tailed significance test*

*$p < .05$

**Role of acceptance (Time 1) in anxiety and depressive symptoms (Time 1 and 2)**

Table 2 summarizes the results of the multiple regression analyses. As none of the socio-demographic and disease-related variables were significantly related to anxiety and depressive symptoms, these variables were not included in the analyses. More acceptance was significantly related to less anxiety symptoms at Time 1 ($\beta$ = -.29, $t$ = -1.86, $p$ = .04, 95% CI [-.51, .02]). Contrary to our expectations, no significant association was found between acceptance at Time 1 and anxiety symptoms at Time 2. Overall, acceptance explained an additional variance of 8% in anxiety symptoms at Time 1 ($F_{change}$ (1, 38) = 3.46, $p$ = .04). Furthermore, more acceptance was significantly related to less depressive symptoms at Time 1 ($\beta$ = -.35, $t$ = -2.33, $p$ = .01, 95% CI [-.33, -.02]), and less depressive symptoms at Time 2 ($\beta$ = -.31, $t$ = -1.71, $p$ = .05, 95% CI [-.56, .05]). Overall, acceptance explained an additional variance of 13% in depressive symptoms at Time 1 ($F_{change}$ (1, 38) = 5.43, $p$ = .01), and 9% in depressive symptoms at Time 2 ($F_{change}$ (1, 25) = 2.92, $p$ =.05). Examination of Tolerance and VIF of all regression analyses revealed no problems of multicollinearity. After controlling for multiple comparisons, all of the initially identified significant associations remained statistically significant, except for the association between acceptance and anxiety symptoms at Time 1 (false discovery rate of 11%) (Benjamini & Hochberg, 1995; Benjamini et al., 2006).

Table 2

*Hierarchical regression analyses of acceptance at Time 1 and anxiety and depression at Time 1 and 2*

| Dependent variable | Step | Predictor | β | R² | ΔR² | Adj. R² |
|---|---|---|---|---|---|---|
| Anxiety (Time 1) | 1 | Acceptance (Time 1) | -.29* | .08* | .08* | .06* |
| Depression (Time 1) | 1 | Acceptance (Time 1) | -.35* | .13* | .13* | .10* |
| Anxiety (Time 2) | 1 | Anxiety (Time 1) | .63* | .39* | .39* | .37* |
| | 2 | Acceptance (Time 1) | .01 | .39 | .00 | .34 |
| Depression (Time 2) | 1 | Depression (Time 1) | .31* | .13* | .13* | .10* |
| | 2 | Acceptance (Time 1) | -.31* | .22* | .09* | .16* |

*One-tailed significance test*

*$p$ < .05*

**Role of acceptance (Time 1) in health-related quality of life (Time 1 and 2)**

Table 3 summarizes the results regarding quality of life. Acceptance was not related to physical functioning at both Time 1 and 2. Furthermore, more acceptance was related to better role functioning ($\beta$ = .32, $t$ = 2.20, $p$ = .02, 95% CI [.11, 2.67]) at Time 1, but not Time 2. Acceptance at Time 1 accounted for an additional variance of 10% in role

functioning at Time 1 ($F_{change}$ (1, 37) = 4.83, $p$ = .02). More acceptance was also related to better emotional functioning ($\beta$ = .34, $t$ = 2.18, $p$ = .02, 95% CI [.10, 2.68]) at Time 1, but not Time 2. Acceptance at Time 1 accounted for an additional variance of 9% in emotional functioning at Time 1 ($F_{change}$ (1, 36) = 4.74, $p$ = .02). Finally, more acceptance was also related to better social functioning ($\beta$ = .48, $t$ = 3.12, $p$ =.00, 95% CI [.60, 2.84]) at Time 1, but not Time 2. Acceptance at Time 1 accounted for an additional variance of 18% in social functioning at Time 1 ($F_{change}$ (1, 37) = 9.70, $p$ = .00). Examination of Tolerance and VIF of all regression analyses revealed no problems of multicollinearity. After controlling for multiple comparisons, all of the initially identified significant associations remained statistically significant (Benjamini & Hochberg, 1995; Benjamini et al., 2006).

Table 3

*Hierarchical regression analyses of acceptance at Time 1 (T1) and physical, role, emotional, and social functioning at Time 1 and 2 (T2)*

| Dependent variable | Step | Predictor | β | R² | ΔR² | Adj. R² |
|---|---|---|---|---|---|---|
| Physical functioning (T1) | 1 | Disease severity | .34* | .10* | .10* | .08* |
| | 2 | Acceptance (T1) | .19 | .14 | .03 | .09 |
| Role functioning (T1) | 1 | Disease severity | .37* | .11* | .11* | .09* |
| | 2 | Acceptance (T1) | .32* | .22* | .10* | .17* |
| Emotional functioning (T1) | 1 | Gender | -.20 | .21* | .21* | .17* |
| | | Time since diagnosis | -.28* | | | |
| | 2 | Acceptance (T1) | .34* | .30* | .09* | .24* |
| Social functioning (T1) | 1 | Gender | -.14 | .12* | .12* | .10* |
| | 2 | Acceptance (T1) | .48* | .31* | .18* | .27* |
| Physical functioning (T2) | 1 | Disease severity | .41* | .29* | .29* | .26* |
| | 2 | Phys. functioning (T1) | .56* | .59* | .31* | .56* |
| | 3 | Acceptance (T1) | .09 | .60 | .01 | .55 |
| Role functioning (T2) | 1 | Disease severity | .26 | .13* | .13* | .09* |
| | 2 | Role functioning (T1) | .32 | .22* | .09* | .16* |
| | 3 | Acceptance (1) | -.01 | .22* | .00* | .12* |
| Emotional functioning (T2) | 1 | Emo. functioning (T1) | .56* | .27* | .27* | .24* |
| | 2 | Acceptance (T1) | -.13 | .29 | .02 | .23 |
| Social functioning (T2) | 1 | Social functioning (T1) | .73* | .56* | .56* | .55* |
| | 2 | Acceptance (T1) | .06 | .57 | .00 | .53 |

*One-tailed significance test*

*$*p < .05$*

**DISCUSSION**

This study is the first to provide insights in the relationship between acceptance and well-being in CF, measured at different time points. The current findings are in line with those from a previous study on acceptance in adolescents and young adults with CF (Casier et al., 2008). This study extends research by incorporating health-related quality of life and investigating the relationships prospectively.

Consistent with previous research (Casier et al., 2008; McCracken & Vowles, 2008; Van Damme et al., 2006), our findings reveal that greater acceptance was related to less depressive symptoms measured at the same point in time, and 6 months later. Contrary to our expectations and to research in chronic pain (see McCracken & Eccleston, 2005), acceptance was not related to anxiety symptoms at Time 1 and 2. This lack of association may most likely be due to our small sample size. It also may reflect the relatively low levels of anxiety symptoms and high levels of acceptance in the current sample. Moreover, it is possible that acceptance is rather related to illness-specific than to general anxiety (McCracken & Eccleston, 2005). The significant relationship between acceptance and depressive symptoms at both Time 1 and 2 is in line with the conceptualisation of acceptance. An important feature of acceptance is to resist the idea that acceptance of illness is a sign of weakness, failure, helplessness, or inferiority (Risdon, Eccleston, Crombez, & McCracken, 2003). Acceptance involves acknowledging the reality of being chronically ill without resigning or surrendering (Evers et al., 2001; Hayes et al., 2006; Risdon et al., 2003). The belief that illness does not imply the end of a meaningful life is indeed inconsistent with depressive symptoms.

Acceptance was also related to several domains of health-related quality of life. More specifically, greater acceptance was related to better role, emotional, and social functioning measured at the same time. This is consistent with research in other chronic conditions such as chronic pain, chronic fatigue syndrome, and multiple sclerosis (Evers et al., 2001; McCracken & Vowles, 2008; Van Damme et al., 2006). Contrary to our expectations, acceptance at Time 1 was not related to physical, role, emotional, and social functioning 6 months later. The most likely explanation for this lack of association may be the small sample size. Despite the lack of association with the Time 2 outcomes, the Time 1 findings are in line with the conceptualisation of acceptance. Acceptance includes broadening a focus upon illness to other domains of life. This engagement in important and achievable developmental goals (e.g., further education, work, hobbies), within the

context of a life-shortening illness, seems a prerequisite for a valued life (Evers et al., 2001; Hayes et al., 2006; Risdon et al., 2003).

The present study contributes to the increasing support that acceptance has positive effects on the adjustment to chronic illness, beyond the effects of socio-demographic and disease-related variables. In the context of CF, acceptance comprises the ability to face the demands and limitations characteristic to CF, and to reconcile to the unpredictable and uncontrollable nature of CF (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003). The current findings indicate that acceptance is related to less depressive symptoms and better health-related quality life at the same time point, and to less depressive symptoms 6 months later. These findings suggest that the role of acceptance may be central during adolescence and young adulthood. Indeed, from adolescence on, CF and its burden typically become more severe (e.g., more frequent symptoms, more exacerbations, more disability, more intensive treatment) (Ernst et al., 2010). This means that the trade-off between CF-related demands and developmental demands (e.g., doing aerosol therapy while studying, learning a new language/reading books/make drawings while hospitalised, choosing a job that fits with one's physical capabilities) becomes essential during this period (Badlan, 2006; Gjengedal et al., 2003; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009). Acceptance may impact this trade-off as it may help adolescents and young adults with CF to keep pursuing important developmental goals (e.g., independence, close relationships, academic achievement, family life, occupation) in the development towards adulthood (Ernst et al., 2010; Gjengedal et al., 2003). The finding that in the current sample acceptance was unrelated to disease severity may indicate that the effects of acceptance rather refer to the subjective experience of CF instead of one's objective medical condition.

Further research is needed to verify whether adolescents and young adults with CF benefit from psychological interventions developed to stimulate acceptance of disease. These interventions may prevent loss of well-being and/or enhance well-being when affected. Approaches that incorporate acceptance, such as Acceptance and Commitment Therapy (ACT; Hayes, 2004; Hayes et al., 2006), already exist for several conditions such as depression, psychosis, cancer, substance abuse, chronic pain, etcetera and reveal promising results (Hayes et al., 2006; Powers, Zum Vörde Sive Vörding, & Emmelkamp, 2009; Vowles, Wetherell, & Sorrell, 2009). Within ACT (Hayes, 2004; Hayes et al., 2006), acceptance is considered as a means of increasing action that is directed toward important life goals, in spite of being ill (Bach & Hayes, 2002; Evers et

al., 2001; Hayes et al., 2006). This may be particularly relevant during adolescence and young adulthood as the ultimate challenge of adolescents and young adults with CF is to balance the pursuit of CF-related goals (i.e., disease management) with the pursuit of other important developmental goals (e.g., independence, close relationships, academic achievement, family life, occupation) (Badlan, 2006; Gjengedal et al., 2003; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009). By acknowledging the reality of having CF without struggle or resignation, by believing that CF does not imply the end of a meaningful life and, by choosing actions that stimulate to strive for important developmental goals despite having CF, a meaningful life and well-being can be sustained (Ernst et al., 2010; Hayes et al., 2006; Risdon et al., 2003). In this context, it may be useful to consider ACT (Hayes, 2004; Hayes et al., 2006) for individuals with CF. However, to determine the usefulness of ACT (Hayes, 2004; Hayes et al., 2006) for adolescents and young adults with CF it is necessary to (a) further investigate the role of acceptance and related processes in the context of CF, (b) identify factors that influence acceptance, (c) modify existing ACT (Hayes, 2004; Hayes et al., 2006) approaches for individuals with CF, (d) conduct feasibility studies, and (e) conduct randomized controlled trials (Gauthier et al., 2009; Glasscoe & Quittner, 2008). Furthermore, as male adolescents/young adults and adolescents/young adults with higher disease severity demonstrated worse outcomes on particular aspects of well-being, special attention should be paid to these adolescents' and young adults' well-being.

There are some limitations to this study. First, the sample size of the present study was small and consequently it is possible that small and medium effects remained undetected. At Time 1, the current study only had sufficient power to detect large effects. At Time 2, power was low, even for the detection of large effects. Furthermore, we controlled for the corresponding Time 1 dependent variable in the regression analyses for the Time 2 outcomes. As the Time 1 dependent variables captured a great amount of the variance in the Time 2 outcomes, this may be another reason for the null findings for anxiety and health-related quality of life at Time 2. Second, part of our findings are cross-sectional and therefore do not indicate causal effects. The prospective part of the current study is an advancement on cross-sectional findings, but only provides a first indication of the long-term effects of acceptance. To truly investigate long-term effects, future research should include measurements at, at least, three consecutive time points. Third, adolescents and young adults reported on average a relatively good well-being. As previous research often reports worse levels of well-being for adolescents and young

adults with CF in comparison with healthy peers (Goldbeck & Schmitz, 2001; Quittner et al., 2008; Pfeffer et al., 2003), it cannot be assumed that the current findings apply to the general population of adolescents and young adults with CF. Further research in more diverse samples is needed to generalise the current findings. Fourth, 40 of 64 adolescents and young adults consented to participate. As this may reflect sample bias, we should be careful in generalising our results to other samples. Of the 24 non-participants, two declined participation because they did not want to be unnecessarily confronted with their CF, what may point to a lack of acceptance. As the mean score for acceptance was quite high in the current sample, it is possible that the current findings may not generalize to samples who score lower on acceptance. Fifth, the questionnaires were administered in two different ways: during a house visit under the supervision of a research assistant at Time 1 and after receiving the questionnaires by mail at Time 2. It is possible that this difference in approach may have affected our results (e.g., low response rate at Time 2). Sixth, acceptance was only measured once. As a consequence, we were unable to detect possible changes in acceptance and to identify if changes in acceptance over time concur with changes in well-being. Seventh, reliabilities for role functioning (Time 2) and social functioning (Time 1) of the CFQ-R Teen/Adult Version were quite low. Therefore, the results for these outcomes should be interpreted with caution. Eighth, acceptance, anxiety and depressive symptoms, and health-related quality of life were only assessed using self-report scales. A multi-method approach (e.g., objective indicators of health-related quality of life, ratings by parents, caregivers, or nurses) would contribute to the generality of the current findings. What concerns acceptance, more instruments should be developed that are usable in individuals with CF or chronic conditions in general, and focus more particular on certain facets of acceptance such as acceptance of difficult thoughts and feelings related to illness and the choice for action driven by important life goals despite being ill. These instruments could be based on the Acceptance and Action Questionnaire (AAQ; Hayes, et al., 2004), a generic measure that assesses ACT processes such as acceptance, values-based action, and psychological flexibility. The AAQ has already been adjusted for specific conditions such as chronic pain (Chronic Pain Acceptance Questionnaire; McCracken, Vowles, & Eccleston, 2004) and diabetes (Acceptance and Action Diabetes Questionnaire; Gregg, 2004, in Hayes et al., 2004), but not yet for CF.

To our knowledge, this is the first prospective analysis of the role of acceptance in anxiety and depressive symptoms, and health-related quality of life of adolescents and young adults with CF. The promising findings from this study indicate that acceptance

may play a protective role in the well-being of these adolescents and young adults. These encouraging results stimulate to replicate the current findings and to investigate other processes related to acceptance such as the pursuit of important developmental goals. The relationship between acceptance and the pursuit of important developmental goals has, to our knowledge, not yet been studied. Future research should address this relationship and should examine if the relationship between acceptance and well-being is mediated by goal pursuit.

**REFERENCES**

Accurso, F. J. (2008). Update in cystic fibrosis 2007. *American Journal of Respiratory and Critical Care Medicine, 177,* 1058-1061. doi: 10.1164/rccm.200801-069UP

Anderson, D.L., Flume, P.A., & Hardy, K.K. (2001). Psychological functioning of adults with cystic fibrosis. *Chest, 119,* 1079-1084. doi: 10.1378/chest.119.4.1079

Bach, P., & Hayes, S.C. (2002). The use of acceptance and commitment therapy to prevent the rehospitalization of psychotic patients: a randomized controlled trial. *Journal of Consulting and Clinical Psychology, 70(5),* 1129-1139. doi: 10.1037//0022-006X.70.5.1129

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Bourke, S.J., Doe, S.J., Gascoigne, A.D., Heslop, K., Fields, M., Reynolds, D., Mannix, K. (2009). An integrated model of provision of palliative care to patients with cystic fibrosis. *Palliative Medicine, 23(6),* 512-517. doi: 10.1177/0269216309106312

Benjamini, Y. & Hochberg, Y. (1995). Controlling the false discovery rate: a practical and powerful approach to multiple testing. *Journal of the Royal Statistical Society, Series B (Methodological), 57(1),* 289-300. Retrieved from www.jstor.org

Benjamini, Y., Krieger, AM, & Yekutieli, D. (2006). Adaptive linear step-up procedures that control the false discovery rate. *Biometrika, 93(3),* 491-507. Retrieved from boimet.oxfordjournals.org

Bregnballe, V., Thastum, M, & Schiotz, P.O. (2007). Psychosocial problems in children with cystic fibrosis. *Acta Paediatrica, 96,* 58-61. doi: 10.1111/j.1651-2227.2006.00014.x

Carefusion (2009). MasterScreen$^{TM}$ Body. Retrieved September 2, 2010, from http://www.carefusion.com/images/MasterScreen_Body_Brochure.pdf

Casier, A., Goubert, L., Huse, D., Theunis, M., Franckx, H., Robberecht, E., Matthijs, D., & Crombez, G. (2008). The role of acceptance in psychological functioning in adolescents with cystic fibrosis: a preliminary study. *Psychology and Health, 23(5),* 629-638. doi: 10.1080/08870440802040269

Cella, D. (1998). Quality of life. In J.C. Holland (Ed.), *Psychooncology* (pp. 1135-1146). New York, Oxford: Oxford University Press.

Cystic Fibrosis Foundation (2005). 2005 annual report. Retrieved July 7, 2008, from http://www.cff.org/aboutCFFoundation/AnnualReport/

Cystic Fibrosis Foundation (2008). Patient registry. Annual Data Report 2008. Retrieved September 2, 2010, from http://www.cff.org/UploadedFiles/research/ClinicalResearch/2008-Patient-Registry-Report.pdf

Cystic Fibrosis Foundation (2010) Living with cystic fibrosis. Retrieved January 21, 2010, from http://www.cff.org/LivingWithCF/

Damme, S. Van, Crombez, G., Houdenhove, B. Van, Mariman, A., & Michielsen, W. (2006). Well-being in patients with chronic fatigue syndrome: The role of acceptance. *Journal of Psychosomatic Research, 61,* 595-599. doi:10.1016/j.jpsychores.2006.04.015

Diener, E., Suh, E.M., Lucas, R.E., & Smith, H.L. (1999). Subjective well-being: three decades of progress. *Psychological Bulletin, 125(2),* 276-302.

Emanuel, E.J., Wendler, D., & Grady, C. (2000). What makes clinical research ethical? *Journal of the American Medical Association, 283,* 2701-2711. Retrieved from www.jama.com

Ernst, M.M., Johnson, M.C., & Stark, L.J. (2010) Developmental and psychosocial issues in cystic fibrosis. *Child & Adolescent Psychiatric Clinics of North America, 19,* 263-283. doi:10.1016/j.chc.2010.01.004

Evers, A.W.M., Kraaimaat, F.W., Lankveld, W. van, Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavorable thinking: The Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Gauthier, L.R., Roding, G., Zimmerman, C., Warr, D., Moore, M., Shepherd, F., & Gagliese, L. (2009). Acceptance of pain: A study in patients with advanced cancer. *Pain, 143,* 147-154. doi:10.1016/j.pain.2009.02.009

Gjengedal, E., Rustøen, T., Wahl, A.K., & Hanestadt, B.R. (2003). Growing up and living with cystic fibrosis. Everyday life and encounters with the health care and social services – A qualitative study. *Advances in Nursing Science, 26 (2)*, 149-159.

Glasscoe, C.A. & Quittner, A.L. Psychological interventions for people with cystic fibrosis and their families. *Cochrane Database of Systematic Reviews* 2008, Issue 3. Art. No.: CD003148. doi: 10.1002/14651858.CD003148.pub2.

Goldbeck, L. & Schmitz, T.G. (2001). Comparison of three generic questionnaires measuring quality of life in adolescents and adults with cystic fibrosis: the 36-item short form health survey, the quality of life profile for chronic diseases, and the questions on life satisfaction. *Quality of Life Research, 10,* 23-36.

Havermans, T., Colpaert, K., & Dupont, L.J. (2008). Quality of life in patients with cystic fibrosis: Association with anxiety and depression. *Journal of Cystic Fibrosis, 7,* 581-584. doi: 10.1016/j.jcf.2008.05.010

Havighurst, J.R. (1971). *Developmental tasks and Education.* New York: Longman.

Hayes, S.C. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapies. *Behavior Therapy, 35,* 639-665. doi: 10.1016/S00005-7894(04)80013-3

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: model, processes and outcomes. *Behaviour Research and Therapy, 44,* 1-25. doi:10.1016/j.brat.2005.06.006

Hayes, S.C., Strosahl, K., Wilson, K.G., Bissett, R.T., Pistorello, J., Toarmino, …McCurry, S.M. (2004). Measuring experiental avoidance: a preliminary test of a working model. *The Psychological Record, 54,* 553-578.

Klijn, P.H., van Stel, H.F., Quittner, A.L., van der Net, J., Doeleman, W., van der Schans, C.P., & van der Ent, C.K. (2004). Validation of the Dutch cystic fibrosis questionnaire (CFQ) in adolescents and adults. *Journal of Cystic Fibrosis, 3,* 29-36. doi:10.1016/j.jcf.2003.12.006

Kline, R. B. (2004). *Beyond Significance Testing: Reforming Data Analysis Methods in Behavioral Research*. Washington DC, WA: American Psychological Association.

Lauwerier, E., Crombez, G., Van Damme, S., Goubert, L., Vogelaers, D., & Evers, A. W. M. (2010). The construct validity of the Illness Cognition Questionnaire: the robustness of the three-factor structure across patients with chronic pain and chronic fatigue. *International Journal of Behavioral Medicine, 17(2),* 90-96. doi: 10.1007/s12529-009-9059-z

Martin P. & Bateson P. (1993, 2007). *Measuring Behaviour: An Introductory Guide*. Cambridge, England: Cambridge University Press.

McCracken, L.M., Vowles, K.E., & Eccleston, C. (2004). Acceptance of chronic pain: component analysis and a revised assessment method. *Pain, 107,* 159-166. doi:10.1016/j.pain.2003.10.012

McCracken, L.M., Carson, J.W., Eccleston, C., Keefe, F.J. (2004) Topical review: acceptance and change in the context of chronic pain. *Pain, 109,* 4-9. doi:10.1016/j.pain.2004.02.006

McCracken, L.M. & Eccleston, C. (2005). A prospective study of acceptance of pain and patient functioning with chronic pain. *Pain, 118,* 164-169. doi:10.1016/j.pain.2005.08.015

McCracken, L.M. & Vowles, K.E. (2008). A prospective analysis of acceptance of pain and values-based action in patients with chronic pain. *Health Psychology, 27(2),* 215-220. doi: 10.1037/0278-6133.27.2.215

McCracken, L.M. & Zhao-O'Brien, J. (2010). General psychological acceptance and chronic pain: There is more to accept than the pain itself. *European Journal of Pain, 14(2),* 170-175. doi:10.1016/j.ejpain.2009.03.004

Pakenham, K.I. (2006). Investigation of the coping antecedents to positive outcomes and distress in multiple sclerosis (MS). *Psychology and Health, 21(5),* 633-649. doi: 10.1080/14768320500422618

Perneger, T.V. (1998). What's wrong with Bonferroni adjustments? *British Medical Journal, 316,* 1236-1238. Retrieved from www.bmj.com

Pfeffer, P.E., Pfeffer, J.M., & Hodson, M.E. (2003). The psychological and psychiatric side of cystic fibrosis in adolescents and adults. *Journal of Cystic Fibrosis, 2,* 61-68. doi:10.1016/S1569-1993(03)00020-1

Powers, M.B., Zum Vörde Sive Vörding, M.B., & Emmelkamp, P.M.G. (2009). Acceptance and Commitment Therapy: A Meta-analytic Review. *Psychotherapy and Psychosomatics, 78,* 73-80. doi: 10.1159/000190790

Quittner, A.L., Barker, D.H., Snell, C., Grimley, M.E., Marciel, K., & Cruz, I. (2008). Prevalence and impact of depression in cystic fibrosis. *Current Opinion in Pulmonary Medicine, 14,* 582-588. doi:10.1097/MCP.0b013e3283121cf1

Quittner, A., Buu, A., Messer, M.A., Modi, A.C., & Watrous, M. (2005). Development and validation of the cystic fibrosis questionnaire in the United States- A health-related quality of life measure for cystic fibrosis. *Chest, 128,* 2347-2354. doi: 10.1378/chest.128.4.2347

Quittner, A.L., Modi, A.C., Watrous, M., & Davis, M.A. (2003). *Cystic Fibrosis Questionnaire Revised (CFQ-R): User's manual.* Washington, DC: Cystic Fibrosis Foundation.

Ramsey, B.W., & Boat, T.F. (1994). Outcome measures for clinical trials in cystic fibrosis. *The Journal of Pediatrics, 124,* 177-192.

Ratjen, F., & Döring, G. (2003). Cystic fibrosis. *The Lancet, 361,* 681-689. Retrieved from www.thelancet.com

Riekert, K.A., Bartlett, S.J., Boyle, M.P., Krishnan, J.A., & Rand, C.S. (2007). The association between depression, lung function, and health-related quality of life among adults with cystic fibrosis. *Chest, 132,* 231-237. doi: 10.1378/chest.06-2474

Risdon, A., Eccleston, C., Crombez, G., & McCracken, L. (2003). How can we learn to live with the pain? A Q-methodological analysis of the diverse understandings of acceptance of chronic pain. *Social Science and Medicine, 56,* 375-386. doi: 10.1016/S0277-9536(02)00043-6

Rosenstein, B.J., & Zeitlin, P.L. (1998). Cystic fibrosis. *Lancet, 351,* 277-282. Retrieved from www.thelancet.com

Schwartz, L.A. & Drotar, D. (2009). Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 34(9),* 954-965. doi:10.1093/jpepsy/jsn144

Snaith, P.R. (2003). The Hospital Anxiety and Depression Scale. *Health and Quality of Life Outcomes, 1,* 29-32. Retrieved from http://www.hqlo.com/content/1/1/29

Spilker (1996). Introduction. In B. Spilker (Ed.), *Quality of life and pharmacoeconomics in clinical trials* (pp. 1-10). Philadelphia, PA: Lippincott-Raven.

Spinhoven, P., Ormel, J., Sloekers, P.P.A., Kempen, G.I.J.M., Speckens, A.E.M., & Hemert, A.M. Van (1997). A validation study of the Hospital Anxiety and Depression Scale (HADS) in different groups of Dutch subjects. *Psychological Medecine, 27,* 363-370. Retrieved from http://journals.cambridge.org

Szyndler, J.E., Towns, S.J., van Asperen, P.P., & McKay, K. (2005). Psychological and family functioning and quality of life in adolescents with cystic fibrosis. *Journal of Cystic Fibrosis, 4,* 135-144. doi:10.1016/j.jcf.2005.02.004

Urquhart Law, G. (2002). Dissimilarity in adolescent and maternal representations of Type 1 diabetes: exploration of relations to adolescent well-being. *Child: Care, Health, & Development, 28 (5),* 369-378.

Vacha-Haase, T. & Thompson, B. (2004) How to estimate and interpret various effect sizes. Journal of Counseling Psychology, 51(4), 473-481. doi: 10.1037/0022-0167.51.4.473

Viane, I., Crombez, G., Eccleston, C., Poppe, C., Devulder, J., Houdenhove, B. Van, & Corte, W. De (2003). Acceptance of pain is an independent predictor of mental well-being in patients with chronic pain: Empirical evidence and reappraisal. *Pain, 106,* 65-72. doi:10.1016/S0304-3959(03)00291-4

Vowles, K. E., Wetherell, J.L., & Sorell, J.T. (2009). Targeting acceptance, mindfulness, and values-based action in chronic pain: findings of two preliminary trials of an outpatient group-based intervention. *Cognitive and Behavioral Practice, 16,* 49-58. Retrieved from www.elsevier.com

Westin, V., Hayes, S.C., & Andersson, G. (2008). Is it the sound or your relationship to it? The role of acceptance in predicting tinnitus impact. *Behaviour Research and Therapy, 46,* 1259-1265. doi:10.1016/j.brat.2008.08.008

White, D., Leach, C., Atkinson, M., & Cottrell, D. (1999). Validation of the Hospital Anxiety and Depression Inventory for use with adolescents. *British Journal of Psychiatry, 175,* 452-454.

White, T., Miller, J., Smith, G.L., & McMahon, W.M. (2009). Adherence and psychopathology in children and adolescents with cystic fibrosis. *European Child & Adolescent Psychiatry, 18(2),* 96-104. doi: 10.1007/s00787-008-0709-5

Wonnacott, R. J. & Wonnacott, T. H. (1985). *Introductory Statistics*. New York, NY: Wiley.

Zigmond, A.S., & Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavia, 67,* 361-370.

# CHAPTER 3

### SPIROMETRY-RELATED PAIN AND DISTRESS IN ADOLESCENTS AND YOUNG ADULTS WITH CYSTIC FIBROSIS: THE ROLE OF ACCEPTANCE[1]

**ABSTRACT**

*Introduction:* Aims of the study were to investigate the occurrence of spirometry-related pain and distress in adolescents and young adults with cystic fibrosis (CF), and the role of acceptance of illness in this pain and distress.

*Method:* Thirty-six adolescents and young adults with CF (12-22 years) completed a questionnaire assessing acceptance. Spirometry-related distress was assessed by self-report (i.e., anxiety/worry about the procedure) and physiological (i.e., heart rate and heart rate variability) outcomes before spirometry. Spirometry-related pain was assessed by self-report (i.e., expected pain and pain-related thoughts). Self-reported distress and pain during spirometry were also assessed.

*Results:* Eighty-nine per cent of the participants reported distress before spirometry, 67% experienced distress during spirometry, 28% expected pain during spirometry, and 22% actually experienced pain. Interestingly, partial correlations revealed that more acceptance was related to less expected pain and pain-related thoughts. Acceptance, however, was unrelated to distress, anxiety, and pain during spirometry.

*Discussion:* The current study indicates that a non-negligible number of adolescents and young adults with CF experience pain and distress during spirometry. Furthermore, results indicate that acceptance may play a protective role in the more indirect consequences of having CF, such as expected pain and pain-related thoughts during medical procedures. Acceptance, however, was not related to distress before and during spirometry, nor to experienced pain. These findings partially contribute to the increasing

support that acceptance may play a protective role in managing the consequences of living with CF.

**INTRODUCTION**

Cystic fibrosis (CF) is the most common chronic genetic condition in the Caucasian population. It affects 1 in 2500 newborn children and results in life-long morbidity and premature mortality (Ratjen & Döring, 2003). CF is characterized by hyper viscous secretions in several organs throughout the body. In the lungs, this mucus triggers recurrent infection, inflammation, and irreversible tissue damage that very often lead to death from respiratory failure (Ratjen & Döring, 2003; Rosenstein & Zeitlin, 1998). In the pancreas, mucus plugging leads to pancreatic insufficiency causing maldigestion and malabsorption of nutrients. As CF progresses, other complications such as diabetes mellitus, osteoporosis, chronic liver disease, and pneumothorax occur (Ratjen & Döring, 2003; Rosenstein & Zeitlin, 1998). This worsening typically occurs during adolescence and may place a serious and increasing burden on affected adolescents and young adults (Ernst, Johnson, & Stark, 2010).

To prevent or delay health damage, a complex and time-consuming daily treatment regimen (i.e., physiotherapy, exercise, nutritional management, aerosol therapy, drug treatment) combined with regular routine hospital visits and, sometimes, hospitalisation is needed (Ayers, Muller, Mahoney, & Seddon, 2011; de Abreu e Silva & Dodge, 1996; O'Sullivan & Freedman, 2009; Ward, Brinkman, Slifer, & Paranjape, 2010). Routine hospital visits include several medical procedures such as weight and height measurement, sputum culture, physical examination, chest X-ray, and spirometry. Undergoing medical procedures may be a distressing experience for children, adolescents, and young adults with CF (Miller et al., 2005; Ward et al., 2010). This might be particularly the case for spirometry, a standard non-invasive procedure for tracking changes in the lungs (Geller & Rubin, 2009). Spirometry entails maximal inhalation followed by forced and maximal exhalation whereby the mouth is placed around a mouthpiece (Miller et al., 2005). Procedure-related distress may be due to experienced discomfort, undue fatigue, and/or dizziness (or even syncope) during performance, but also difficulty with the technical performance of the manoeuvres, and intolerance of the procedure due to substantial and ongoing anxiety and/or worry with health status and well-being (Davis & Alton, 2009; Geller & Rubin, 2009; Miller et al., 2005; Ward et al., 2010). Further, spirometry may not only evoke distress, but also pain (e.g., chest pain) (Ernst et al., 2010; Miller et al., 2005). Given the possibility of pain and distress during spirometry, the potential detrimental effects of pain and distress, and the fact that spirometry is an important medical procedure that individuals with CF must regularly

undergo, it is important to verify whether spirometry indeed evokes considerable levels of pain and distress (Geller & Rubin, 2009; Miller et al., 2005; Ward et al., 2010). To the best of our knowledge, the occurrence of spirometry-related pain and distress has yet to be investigated.

Extensive research in a variety of invasive and non-invasive medical procedures has shown that assessing and understanding procedural pain and distress in adolescents and young adults with chronic illness is essential as this can interfere with cooperation during the procedure, cause memory and attentional biases towards negative aspects of the procedure, influence the management of subsequent medical procedures, cause persistent conditioned distress, and negatively impact upon quality of life (Chen, Zeltzer, Craske, &Kask, 2000; Klosky et al., 2007; Tucker, Slifer, & Dahlquist, 2001). Adequate management of procedure-related pain and distress, which may involve pharmacological (e.g., analgesics) as well as non-pharmacological interventions (e.g., distraction, cognitive-behavioral interventions), is therefore of utmost importance (Ayers et al., 2011; Ernst et al., 2010). Previous research has identified several risk factors that contribute to increased pain and distress during medical procedures such as younger age, female gender, negative memory, and higher expectations of pain and distress (Klosky et al., 2007; Rudolph, Dennig, & Weisz, 2005; Tucker et al., 2001). However, it is equally important to gain insight into the factors that *protect* against pain and distress. Understanding these protective factors may guide the prevention of pain and distress during medical procedures and/or attenuate the short and long term negative consequences of pain and distress. To date, only a few studies have investigated factors that may protect against procedure-related pain and/or distress. Protective factors that have been identified include the use of control, distraction, and humor (Ayers et al., 2010; Schechter et al., 2007).

Another potentially protective factor for procedure-related pain and distress is acceptance. Acceptance can be defined as "recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its aversive consequences" (Evers et al., 2001). Research into the role of acceptance, conducted across several paediatric chronic conditions such as chronic pain, juvenile arthritis, and sickle cell disease, has revealed that greater acceptance is related to better overall functioning. Specifically, higher acceptance has been associated with less anxiety, less depression, less disability, and a better quality of life (e.g., Feinstein et al., 2011; Masuda, Cohen, Wicksell, & Kenmani, 2011; McCracken,

Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olsson, & Hayes, 2011). To date only two studies, reporting the same associations, specifically studied acceptance in adolescents and young adults with CF (Casier et al., 2008, 2011). Furthermore, research in non-clinical samples has demonstrated that higher acceptance was related to less negative emotions during negative experiences (Hofmann, Heering, Sawyer, & Asnaani, 2009; Shallcross, Troy, Boland, & Mauss, 2010).

To our knowledge, there are no studies available that have assessed the impact of acceptance upon procedure-related distress and pain in the context of CF. The present study therefore aimed, within a sample adolescents and young adults with cystic fibrosis, at exploring the occurrence of spirometry-related pain and distress as assessed by self-report and physiological outcomes, and to investigate the explanatory value of acceptance in this pain and distress. In line with previous research on acceptance, it was hypothesized that greater acceptance is related to less expected/experienced pain before/during spirometry and to lower levels of distress before/during spirometry.

**METHOD**

**Participants**

Adolescents and young adults with cystic fibrosis (age range 12-22) were recruited from the University Hospitals Antwerp, Brussels, Ghent, and the Sint-Vincentius Hospital, Antwerp, Belgium. To be eligible, participants had to meet the following criteria: (1) understanding the Dutch language, (2) no developmental disorder, and (3) no (planned) lung transplantation. Adolescents and young adults who had/were awaiting lung transplantation were not included as they find themselves in an extreme situation that is very different from the situation of individuals in the pre-transplant period (e.g., very high level of symptoms, being terminally ill, intertwinement of end of life issues and hope of transplantation). Seventy-eight adolescents and young adults were invited to participate either by letter and/or approached during routine clinic visits by the psychologist from the respective hospital (time period of recruitment: 13 months). Of the 78 adolescents and young adults who were invited, 52 agreed to participate and were contacted by phone by a research assistant (response rate = 66.67%). The main reasons for not participating were a lack of time, lack of motivation, being overloaded with participating in scientific research, or not wanting to be unnecessarily confronted with the disease. Twelve adolescents and young adults could not be enrolled in the study due to the absence of any hospital visit during the period of recruitment. Of the 40 adolescents

and young adults who participated in our study, four were excluded because their observed spirometry procedure deviated from the standard spirometry procedure. The final sample consisted of 36 adolescents and young adults with CF (17 male; 47.22%). Participants' mean age was 17.58 years ($SD = 3.25$). All adolescents and young adults were from Caucasian origin. To determine disease severity, the US CF Foundation guidelines for severity of lung disease were used (Cystic Fibrosis Foundation, 2008). Mean disease severity reflected mild lung disease ($M_{FEV1\%} = 84.27\%$). Mean time since diagnosis was 15.62 years ($SD = 4.48$).

**Procedure**

This study consisted of two parts. The first part consisted of a home visit whereby written parent consent and/or adolescent assent/consent was obtained and all adolescents and young adults were requested to fill out a battery of questionnaires, among which a measure of acceptance. The second part took place during the first hospital visit following the home visit (time range between home and hospital visit = 1-254 days, $M = 65$ days, $SD = 64$ days). During this hospital visit, spirometry-related pain and distress were assessed by means of self-report and physiological measurement (i.e., heart rate). After filling out the self-report measures on pain and distress, a Polar heart rate monitor was placed and set to work while the participants were seated in a quiet place. Subsequently, the spirometry procedure was performed using a Jaeger MasterScreen TM Body (CareFusion Germany 234 GmbH, Hoechberg, Germany). During spirometry, individuals were seated in a glass cabin where they performed maximal inspiratory and expiratory manoeuvres through a mouthpiece. After the spirometry procedure, self-reported distress and pain during spirometry were assessed. This study was approved by the ethical committees of the University Hospital, Antwerp, Brussels, and Ghent, and the Sint-Vincentius Hospital Antwerp and was carried out in accordance with universal ethical principles (Emanuel, Wendler, & Grady, 2000).

**Measures**

*Acceptance* was assessed by means of a Dutch version of the acceptance subscale of the Illness Cognition Questionnaire (ICQ; Evers et al., 2001; Lauwerier et al., 2010). Acceptance is conceptualised as the perceived ability to live with the illness and to master its negative consequences (e.g., "I can handle the problems related to my illness"). This subscale consists of six items, rated on a 4-point Likert scale (1 = *not at all*, 4 = *completely*). Item scores are summed, resulting in a total score ranging from 6 to 24 with higher scores indicating higher levels of acceptance. Psychometric research in samples of

adults with rheumatoid arthritis, multiple sclerosis, chronic pain, and chronic fatigue syndrome has demonstrated adequate reliability and validity of the ICQ (Evers et al., 2001; Lauwerier et al., 2010). Reliability of the acceptance scale in this sample was excellent (Cronbach's $\alpha$ = .90).

*Pain* was assessed before and after spirometry. Participants were requested to rate how much pain they expected to have/they had experienced during spirometry ("How much pain do you think you will have/did you have during the spirometry"; 0 = *no pain at all*, 10 = *a lot of pain*). Scores of 1–3 were considered to indicate mild pain, scores of 4–6 moderate pain, and scores of 7–10 severe pain (Bouhassira, Lantéri-Minet, Attal, Laurent, & Touboul, 2008).

Furthermore, *thoughts related to pain* before spirometry were assessed with a spirometry-specific measure of the Situational Catastrophizing Scale (Campbell et al., 2010a; Campbell, Quartana, Buenaver, Haythornthwaite, & Edwards, 2010b). In line with earlier research (Caes, Vervoort, Eccleston, Vandenhende, & Goubert, 2011; Caes, Vervoort, Trost, & Goubert, 2012; Goubert, Vervoort, Cano, & Crombez, 2009), the spirometry-specific Situational Catastrophizing Scale consisted of one adapted item from each subscale (Rumination: "At this moment, to what extent do you keep thinking about how much pain the spirometry measurement could cause?"; Magnification: "At this moment, to what extent do you think that, because of the pain, something serious could happen to you during the spirometry measurement?"; Helplessness: "At this moment, to what extent do you think that there is nothing you can do to stop the pain you will feel during the spirometry measurement?"). Use of situation-specific measures of catastrophizing is in line with earlier research and recently was explicitly recommended (Campbell et al., 2010a, 2010b). Although repeatedly used, the 3-item version of the scale has not yet been validated. Items were rated on an 11-point Likert scale (0 = *not at all*, 10 = *a lot*) and summed to obtain a total score. This total score varied between 0 and 30, with higher scores indicating higher catastrophizing about pain. Cronbach's $\alpha$ for the spirometry-specific PCS was .89.

*Distress* was assessed by means of self-report and physiological recording of heart rate (variability) before and after spirometry. Before spirometry assessment, participants were requested to rate (a) how anxious/tense they felt ("How anxious/tense do you feel about the spirometry"; 0 = *not anxious/tense at all*, 10 = *very anxious/tense*), (b) how worried they were about the result of spirometry ("How worried are you about the results of the spirometry"; 0 = *not worried at all*, 10 = *very worried*), and (c) how

worried they were about the treatment subsequent to spirometry ("How worried are you about the treatment that will follow upon the spirometry measurement"; 0 = *not worried at all*, 10 = *very worried*). After the spirometry procedure, the level of self-reported distress during spirometry was assessed ("How anxious/tense did you feel during spirometry"; 0 = *not anxious/tense at all*, 10 = *very anxious/tense*). Scores of 1–3 were considered to indicate mild distress or worry, scores of 4–6 moderate distress or worry, and scores of 7–10 severe distress or worry (Bouhassira et al., 2008). Physiological distress before spirometry was measured by means of heart rate and heart rate variability. Beat-to-beat fluctuations were continuously monitored by a wrist heart rate monitor with electrocardiogram precision (Polar S810$^{TM}$, Polar Electro Oy, Kempele, Finland). Heart rate recording started at least three minutes before the start of the spirometry procedure and continued until spirometry measurement was finished. As both the manoeuvres performed during spirometry and the use of Ventolin® (i.e., a bronchodilator inhaled during the spirometry procedure; GlaxoSmithKline BV, Zeist, The Netherlands), impact upon physiological measures, only heart rate data recorded before the spirometry procedure were used. Using a stopwatch, a research assistant noted a timeline of events e.g., baseline monitoring, entering spirometry cabin, start spirometry). A sequence of one minute preceding the start of the spirometry measurement and a baseline sequence of one minute during which the participants were seated in a calm room were extracted from the continuous heart rate recording for further analysis (Polar Precision Performance$^{TM}$ Software, Polar Electro Oy, Kempele, Finland). Measurement errors were filtered with a moderate filter power and a minimum protection zone of six beats per minute. To determine the physiological stress responses (heart rate and heart rate variability), these sequences were further analyzed with HRV Analysis Software (Biomedical Signal Analysis Group Department of Applied Physics, University of Kuopio, Finland). Averages of heart rate and the root mean square of successive differences of inter beat intervals (RMSSD) were obtained. Previous research has shown that RMSSD is a reliable measure of heart rate variability. Higher heart rate indicates higher distress, whereas higher RMSSD is indicative of lower distress (Camm et al., 1996).

*Disease severity* was determined by using forced expiratory volume in one second (FEV$_1$% predicted) measured during the spirometry procedure performed at the day of the hospital visit. FEV$_1$% predicted was used as a continuous variable, with lower values indicating poorer respiratory functioning and more lung disease.

*Socio-demographic information* (age, gender and time since diagnosis) was verbally inquired before administration of the questionnaires.

**Statistical analyses**

All statistical analyses were performed using SPSS 15.0. First, some preliminary analyses were performed. Tests of normality (i.e. Kolmogorov-Smirnov test) were performed for all continuous variables. Descriptive statistics (means, *SD*'s, Cronbach's α) were computed for all variables of interest. Correlational analyses were performed to determine whether age, time since diagnosis, disease severity and acceptance were related to the pain and distress measures. Pearson correlations were performed in the case of normal distribution, and Spearman correlations in the case of non-normal distribution. Furthermore, *t*-tests were performed to examine gender differences for pain and distress.

Separate hierarchical regression analysis (for normal distributed outcomes) or partial/Spearman correlations (for non-normal distributed outcomes) were used to investigate the explanatory value of acceptance for distress and pain. Sociodemographic and disease-related variables that were significantly related to the outcome variables were entered as control variables in a first step. Heart rate and heart rate variability outcomes prior to spirometry were also controlled for baseline heart rate respectively baseline heart rate variability in a second step. Acceptance was entered in the last step.

The current sample had sufficient power (.80) to detect large effects in multiple regressions ($f^2$ = .35) with three variables (Casier et al., 2011; Cohen, 1992).

**RESULTS**

Tests of normality (i.e., Kolmogorov-Smirnov tests) revealed that the scores for the distress outcomes (self-reported and physiological) were normally distributed. Scores for expected pain, pain-related thoughts, and experienced pain, however, were non-normally distributed. Inspection of skewness and kurtosis indicated these variables were skewed to the right. Log transformation of skewed variables did not result in normal distribution for these variables. Therefore, partial correlations (in case of significant control variables) or Spearman correlations (in case of non-significant control variables) (see Table 1) were used to investigate the role of acceptance in expected pain and pain-related thoughts.

**Descriptive statistics**

Means, standard deviations, internal consistencies (Cronbach's α) of all variables, and correlations between age, time since diagnosis, disease severity, acceptance, and the

distress and pain variables are reported in Table 1. Disease severity ranged from not severe/normal lung function ($FEV_1\%$ predicted $\geq 90\%$) to severe ($FEV_1\%$ predicted $< 40\%$). Of all adolescents and young adults, 38.9% had normal lung function ($FEV_1\%$ predicted $\geq 90\%$), 44.4% mild lung disease ($89\% \geq FEV_1\%$ predicted $\geq 70\%$), 13.9% moderate lung disease ($69\% \geq FEV_1\%$ predicted $\geq 40\%$), and 2.8% had severe lung disease ($FEV_1\%$ predicted $< 40\%$). Of the sociodemographic and disease-related variables, only age was significantly related to heart rate prior to spirometry ($r = .35$, $p < .05$). Furthermore, $t$-tests revealed that gender was significantly related to anxiety/tension prior to spirometry ($M_{male} = 1.47$, $SD_{male} = 2.50$; $M_{female} = 3.53$, $SD_{female} = 2.39$; $F(1,34) = 6.35$, $p = .02$; *Cohen's d* = 0.84), worry about results of spirometry ($M_{male} = 2.71$, $SD_{male} = 2.54$; $M_{female} = 5.05$, $SD_{female} = 2.80$; $F(1,34) = 6.87$, $p = .01$; *Cohen's d* = 0.88), pain-related thoughts ($M_{male} = 0.71$, $SD_{male} = 1.72$; $M_{female} = 2.89$, $SD_{female} = 4.05$; $F(1,34) = 4.26$, $p = .04$; *Cohen's d* = 0.70), and acceptance ($M_{male} = 19.94$, $SD_{male} = 3.40$; $M_{female} = 16$, $SD_{female} = 4.20$; $F(1,34) = 9.42$, $p = .00$; *Cohen's d* = 1.03), with females scoring higher on anxiety prior to spirometry, worry about results of spirometry, and pain-related thoughts and lower on acceptance than males.

Table 1

*Means (M), standard deviations (SD), internal consistency (Cronbach's α) and Pearson product-moment/Spearman correlations of age, time since diagnosis, FEV$_1$% predicted, acceptance, anxiety/tension prior to/during spirometry, worry about results of spirometry, worry about treatment subsequent to spirometry, heart rate, heart rate variability, expected and experienced pain during spirometry, and pain-related thoughts*

| | M | SD | α | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1. Age | 17.58 | 3.25 | - | .71** | -.36* | .10 | .06 | .05 | .21 | -.35* | .26 | .02 | -.07 | .02 | .13 |
| 2. Time since diagnosis | 15.62 | 4.48 | - | - | -.35* | .02 | .03 | .07 | .08 | -.31 | .28 | .18 | .06 | .09 | .24 |
| 3. FEV$_1$% predicted | 84.27 | 17.95 | - | - | - | -.11 | -.27 | -.16 | -.04 | .08 | -.29 | .03 | .19 | -.24 | -.13 |
| 4. Acceptance | 17.86 | 4.28 | .90 | - | - | - | -.02 | -.13 | -.14 | .21 | -.22 | -.41* | -.35* | -.02 | -.11 |
| 5. Anxiety/tension before spirometry | 2.56 | 2.62 | - | - | - | - | - | .70** | .64** | .02 | .12 | .22 | -.06 | .91** | .34* |
| 6. Worry results spirometry | 3.94 | 2.90 | - | - | - | - | - | - | .66** | .01 | .09 | .24 | .09 | .67** | .39* |
| 7. Worry treatment spirometry | 3.11 | 2.84 | - | - | - | - | - | - | - | -.13 | .19 | .36* | .26 | .60** | .43* |
| 8. Heart rate | 99.26 | 13.10 | - | - | - | - | - | - | - | - | -.54** | -.26 | -.19 | -.07 | -.19 |
| 9. Heart rate variability | 27.85 | 12.83 | - | - | - | - | - | - | - | - | - | .18 | -.05 | .16 | .19 |
| 10. Expected pain during spirometry | .55 | .97 | - | - | - | - | - | - | - | - | - | - | .50** | .13 | .42* |
| 11. Worrisome pain-related thoughts | 1.86 | 3.32 | - | - | - | - | - | - | - | - | - | - | - | .43** | .35* |
| 12. Anxiety/tension during spirometry | 2.25 | 2.52 | - | - | - | - | - | - | - | - | - | - | - | - | .37* |
| 13. Experienced pain during spirometry | .58 | 1.27 | - | - | - | - | - | - | - | - | - | - | - | - | - |

*Note.* Correlations regarding expected and experienced pain are Spearman correlations; correlations regarding pain-related thoughts are partial correlations controlled for gender; *p < .05; **p < .01

**Prevalence rates of spirometry-related pain and distress**

In general, 27.8% of all participating adolescents and young adults expected some amount of pain during spirometry. The mean level of expected pain was mild, with an average of 0.55 ($SD$ = .97; median = 0; observed range 0-3). No participants expected moderate or severe pain during spirometry. The mean level of pain-related thoughts was rather low ($M$ = 1.86; $SD$ = 3.32; $Md$ = 0; observed range 0-10). About 67% of all adolescents and young adults reported experienced anxiety and/or tension prior to spirometry ($M$ = 2.56; $SD$ = 2.62; $Md$ = 2; observed range 0-9). Specifically, 38.9% of the adolescents and young adults experienced mild anxiety and/or tension prior to spirometry, 16.7% experienced moderate anxiety and/or tension, and 11.1% experienced severe anxiety and/or tension. Further, 80.6% of the participants reported worries about the results of spirometry ($M$ = 3.94; $SD$ = 2.90; $Md$ = 3.5; observed range 0-10), with 30.6 % reporting mild worry, 30.6% moderate worry, and 19.4% severe worry. Finally, 75% of the adolescents and young adults reported worry about the treatment subsequent to spirometry ($M$ = 3.11; $SD$ = 2.84; $Md$ = 2.5; observed range 0-9), with 36.1% reporting mild worry, 25% moderate worry, and 13.9% severe worry.

After spirometry, 22.2% of all participants reported to have actually experienced pain during spirometry ($M$ = 0.58; $SD$ = 1.27; $Md$ = 0; observed range 0-5; mild pain: 16.6%; moderate pain: 5.6%; severe pain: 0%). Furthermore, 61.1% reported to have experienced anxiety and/or tension during spirometry ($M$ = 2.25; $SD$ = 2.52; $Md$ = 1.5; observed range 0-9; mild anxiety/tension: 33.3%; moderate anxiety/tension: 22.2%; severe anxiety/tension: 5.6%).

**Role of acceptance in spirometry-related pain**

Results indicated that more acceptance was related to less expected pain ($rs$ = -.41, $p$ = .02) and fewer pain-related thoughts ($rs$ = -.35, $p$ = .03). The explained variance in expected pain and pain-related thoughts, however, was small (17% and 13% respectively). Acceptance was not related to experienced pain during spirometry ($rs$ = -.11, $p$ = .50) (see Table 1).

**Role of acceptance in spirometry-related distress**

Gender and age were entered as control variables because both variables showed significant associations with distress. Findings indicated that acceptance was not related to anxiety and/or tension prior to and during spirometry, nor to physiological distress and to worry about the results of and treatment subsequent to spirometry (see Table 2).

Examination of Tolerance and variance inflation factors (VIF) of all regression analyses revealed no problems of multicollinearity.

Table 2

*Hierarchical regression analyses of acceptance and anxiety/tension prior to/during spirometry, worry about results of spirometry, worry about treatment subsequent to spirometry, heart rate (HR), and heart rate variability (HRV)*

| Dependent variable | Step | Predictor | β | $R^2$ | $\Delta R^2$ | Adj. $R^2$ |
|---|---|---|---|---|---|---|
| Anxiety/tension prior to spirometry | 1 | Gender | .49** | .16* | .16* | .13* |
| | 2 | Acceptance | .21 | .19 | .03 | .14 |
| Worry about results | 1 | Gender | .45* | .17* | .17* | .14* |
| | 2 | Acceptance | .08 | .17 | .00 | .12 |
| Worry about treatment | 1 | Acceptance | -.13 | .02 | .02 | -.01 |
| HR | 1 | Age | -.23 | .12* | .12* | .10* |
| | 2 | HR baseline | .60** | .49** | .37** | .46** |
| | 3 | Acceptance | .17 | .52 | .03 | .47 |
| HRV | 1 | HRV baseline | .54** | .30** | .30** | .29** |
| | 2 | Acceptance | -.19 | .34 | .04 | .30 |
| Anxiety/tension during spirometry | 1 | Acceptance | -.02 | .00 | .00 | -.03 |

*$p < .05$, **$p < .01$

## DISCUSSION

To our knowledge, this is the first prospective study investigating spirometry-related pain and distress in adolescents and young adults with cystic fibrosis, and the explanatory value of acceptance in this pain and distress. The present findings are the first to indicate that some adolescents and young adults indeed expect and/or experience pain and/or distress during spirometry. Of these adolescents and young adults a non-negligible number reported considerable pain and distress. It should however be noted that, as a group, adolescents and young adults with CF only reported mild expected and experienced pain, low levels of pain-related thoughts prior to spirometry, mild to moderate distress prior to spirometry, and mild experienced distress during spirometry. Although acceptance was unrelated to anxiety and/or tension prior to/during spirometry, worry about the results of spirometry, worry about treatment subsequent to spirometry, physiological distress (heart rate and heart rate variability) prior to spirometry, and experienced pain during spirometry, findings revealed that higher acceptance of illness did relate to lower levels of expected pain and less pain-related thoughts. Given the mild

levels of expected and experienced pain in the current sample and the small portion of explained variance by acceptance, these relationships need to be interpreted with care.

The current findings are the first to provide insight in the occurrence of pain and distress prior to/during spirometry in adolescents and young adults with CF and the explanatory value of acceptance for pain and distress. Investigating procedure/spirometry-related pain and distress is essential as both pain and distress may have a negative impact upon the experience of (subsequent) spirometry and upon quality of life in general (Chen et al., 2000; Klosky et al., 2007; Tucker et al., 2001). The occurrence of pain and distress in the current study is more or less in line with existing literature assuming that spirometry may be painful and/or distressing for adolescents and young adults with CF (Davies & Alton, 2009; Miller et al., 2005). Given these prevalence rates, it can be argued that special attention should be paid to the way adolescents and young adults experience routine spirometry. In this way, possible pain and distress can be adequately detected and managed (e.g., analgesics, distraction, cognitive-behavioural interventions, etc.) when necessary, and short and long term negative consequences of distress and pain (e.g., anxiety, decreased quality of life) can be attenuated. This may be particularly the case for female adolescents and young adults as they reported more anxiety prior to spirometry, more worry about results of spirometry, more pain-related thoughts, and less acceptance than males. Future research, however, is needed to replicate the findings of the current study concerning the occurrence of pain and distress during spirometry in male/female adolescents and young adults with CF. This research should also incorporate measures of quality of life to gain insight in the long term consequences of this pain and distress.

The current study also revealed that acceptance of illness may have the potential to serve as a protective factor for anticipated pain during spirometry and worrisome pain-related thoughts. These findings are, to some extent, consistent with results of previous research. For example, research in adults with chronic pain has reliably demonstrated that acceptance is associated with lower levels of pain-related distress, pain-related anxiety and worrisome thoughts about pain (McCracken & Vowles, 2008; McCracken, Vowles, & Eccleson, 2004; Vowles, McCracken, & Eccleston, 2008). The current study, which is to the best of our knowledge the first that assessed the role of acceptance for acute pain, partially corroborates these previous findings by demonstrating that acceptance may also be related to aspects of acute pain, such as expected pain and pain-related thoughts prior to spirometry. However, and contrary to previous research in chronic pain (McCracken,

& Vowles, 2008), the relationship between acceptance and experienced acute pain did not reach significance within the present study.

Similarly, no evidence was obtained for the role of acceptance in spirometry-related distress (self-report and physiological measures). This is, again, contrary to previous research in paediatric chronic conditions such as chronic pain, juvenile arthritis, cystic fibrosis, and sickle cell disease demonstrating that acceptance is inversely related to several aspects of distress in chronic illness such as positive and negative mood, anxiety, and depression (Casier et al., 2008, 2011; Feinstein et al., 2011; Masuda, Cohen, Wicksell, & Kenmani, 2011; McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olsson, & Hayes, 2011). The current findings are also not in line with research in non-clinical samples showing that higher acceptance is related to less negative emotions (e.g., stress, negative affect) during negative emotional experiences (Hofmann et al., 2009; Shallcross et al., 2010). Although this may suggest that acceptance primarily relates to pain and distress in the context of chronic pain rather than acute pain, further research is warranted to investigate whether acceptance is important in understanding single and repeated episodes of acute pain (i.e., recurrent painful medical procedures) and associated distress. One possible explanation for the lack of association between acceptance and subjective distress/pain is that the spirometry procedure was, at least for the majority of participants, insufficiently distressing/painful to elicit the beneficial effects of acceptance (Shallcross et al., 2010). Further, with respect to the physiological assessment of distress, it may be that increased heart rate, a common symptom in individuals with CF, masked the possible occurrence of subtle increases in heart rate due to physiological stress (National Heart Lung and Blood Institute, 2009). However, it also remains possible that insufficient power to detect small and medium effects underlies the absence of a significant association between acceptance and acute experienced pain and distress. Regarding the psychophysiological measures, it is possible that acceptance is not related to heart rate prior to spirometry, but to the amount of heart rate recovery after the experience of distress. This effect could, however, not be examined in the current study as performing spirometry has, in itself, an impact upon heart rate (i.e., effects of Ventolin® and respiratory manoeuvres).

To some extent, the current findings add to the notion that acceptance may play a protective role in the adjustment to living with chronic illness in general, and CF in particular. In the context of CF, acceptance comprises the ability to face the demands and limitations characteristic to CF, and to reconcile to the unpredictable and uncontrollable

nature of CF (Evers et al., 2001; Badlan, 2006; Gjengedal, Rustøe, Wahl, & Hanestad, 2003). The current findings cautiously indicate that acceptance of illness may also comprise the ability to face more indirect demands of having CF, such as the need to perform spirometry. Although acceptance of illness concerns adjustment to illness in general, the current findings suggest the likelihood that it also impacts upon very specific illness-related experiences such as expected pain during spirometry and pain-related thoughts. From adolescence on, CF typically becomes more severe and consequently requires more intensive medical follow up (Ernst et al., 2010). Therefore, acceptance of CF may be particularly important during adolescence. Approaches addressing acceptance, such as Acceptance and Commitment Therapy (ACT; Hayes, Luoma, Bond, Masuda, Lillis, 2006) already exist for several chronic conditions (e.g., depression, psychosis, Type 2 diabetes, sickle cell disease, epilepsy, chronic pain, and substance abuse) and are promising (Hayes, 2004; Hayes, Luoma, Bond, Masuda, Lillis, 2006; Masuda et al., 2011; Wicksell et al., 2011; Wicksell, Dahl, Magnusson, & Olsson, 2005; Wicksell, Melin, Lekander, & Ollson, 2009). Further research into the role of acceptance in the experiences of adolescents and young adults with CF during painful and distressing medical procedures is needed to determine if these interventions may prove useful for this population.

Procedure-related pain and distress in adolescents and young adults with CF has received little attention thus far. The present findings are among the first of their kind and are likely a first step towards more research on this topic. However, some limitations need to be considered, each of which pointing to directions for future research. First, the study only had sufficient power to detect large effects. It is therefore -as noted earlier- possible that small and medium effects remained undetected. Larger samples are needed to detect smaller effects and to investigate whether gender moderates the effects of acceptance. Second, adolescents and young adults reported, on average, low levels of distress and pain. It is possible that our results do not generalize to adolescents and young adults experiencing high pain and distress. Future research, including other physiological measures (e.g., blood pressure and respiratory rates), is needed to address the impact of acceptance for more distress evoking medical procedures such as venepuncture. Third, only 52 of 78 adolescents and young adults consented to participate, potentially reflecting sample bias. Because mean acceptance was quite high in the current sample, it is possible that the current findings do not generalize to samples scoring lower on acceptance. Future research is needed to replicate the current findings in larger and more diverse (e.g., also

adolescents and young adults who report severe pain, severe distress, and low acceptance) samples. Fourth, there was a long time delay ($M$ = 65 days) between the self-report of acceptance and the report of pain and distress measured during the hospital visit. This interval varied substantially between participants ($SD$ = 64 days) and may have influenced the current results (e.g., effect of intermediate factors). Fifth, pain catastrophizing was measured with three and distress with a single item. Single or low numbers of items may decrease statistical power to detect differences, and are less reliable. Future research may include more comprehensive measures. Sixth, disease severity was only assessed by $FEV_1$% predicted. Use of more complex indexes, including all aspects of disease (see Liou et al., 2001) may provide a more complete image of one's health status. Finally, our measure of acceptance does not fully capture the richness of the construct of acceptance as it has been developed in scientific and clinical use. To be able to assess the very specifics of acceptance in individuals with CF a measure of acceptance that focuses on the specific characteristics of CF and covers all aspects of acceptance is needed. This measure should, for example, assess the ability to face the demands and limitations typical for CF, the capability of letting go the struggle for change or control, reconciliation with the unpredictable and uncontrollable nature of CF. The Acceptance and Action Questionnaire (AAQ) (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003; Heyes et al., 2006) could be particularly informative in this regard. The AAQ is a generic measure that assesses ACT processes such as acceptance, values-based action, and psychological flexibility. The AAQ has already been adjusted for specific conditions such as chronic pain (Chronic Pain Acceptance Questionnaire; 37) and diabetes (Acceptance and Action Diabetes Questionnaire; Hayes et al., 2006), but not yet for CF.

**REFERENCES**

Ayers, S., Muller, I., Mahoney, L., & Seddon, P. (2011). Understanding needle-related distress in children with cystic fibrosis. *British Journal of Health Psychology, 16,* 329-343. doi: 10.1348/135910710X506895

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Bouhassira, D., Lantéri-Minet, M., Attal, N., Laurent, B., & Touboul, C. (2008). Prevalence of chronic pain with neuropathic characteristics in the general population. *Pain, 136,* 380-387. doi: 10.1016/j.pain.2007.08.013

Caes, L., Vervoort, T., Eccleston, E., Vandenhende, M., & Goubert, L. (2011). Parental catastrophizing about child's pain and its relationship with activity restriction: the mediating role of parental distress. *Pain, 152,* 212-222. doi: 10.1016/j.pain.2010.10.037

Caes, L., Vervoort, T., Trost, Z., & Goubert, L. (2012). Impact of parental catastrophizing and contextual threat on parents' emotional and behavioral responses to their child's pain. *Pain, 153,* 687-695.

Camm, A.J., Malik, M., Bigger, B.T., Breithardt, G., Cerutti, S., Cohen, R.J., …Singer, D.H. (1996). Heart rate variability. Standards of measurement, physiological interpretation, and clinical use. *European Heart Journal, 17(3),* 354-381.

Campbell, C.M., Kronfli, T., Buenaver, L.F., Smith, M.T., Berna, C., Haythornthwaite, J.A., & Edwards, R.R. (2010a). Situational versus dispositional measurement of catastrophizing: associations with pain responses in multiple samples. *The Journal of Pain, 11,* 443-453. doi:10.1016/j.jpain.2009.08.009

Campbell, C.M., Quartana, P.J., Buenaver ,L.F., Haythornthwaite, J.A., & Edwards, R.R. (2010b). Changes in situation-specific pain catastrophizing precede changes in pain report during capsaicin pain: a cross-lagged panel analysis among healthy, pain-free participants. *The Journal of Pain, 11,* 876-884. doi:10.1016/j.jpain.2009.12.007

Casier, A., Goubert, L., Huse, D., Theunis, M., Franckx, H., Robberecht, E., Matthijs, D., & Crombez, G. (2008). The role of acceptance in psychological functioning in adolescents with cystic fibrosis: a preliminary study. *Psychology and Health, 23(5),* 629-638. doi: 10.1080/08870440802040269

Casier, A., Goubert, L., Theunis, M., Huse, D., De Baets, F., Matthys, D., & Crombez, G. (2011). Acceptance and well-being in adolescents and young adults with cystic

fibrosis: a prospective study. *Journal of Pediatric Psychology, 36,* 476-487. doi: 10.1093/jpepsy/jsq111

Chen, E., Zeltzer, L.K., Craske, M.G., & Katz, E.R. (2000). Children's memories for painful cancer treatment procedures: implications for distress. *Child Development, 71,* 931-945.

Cohen J. (1992). A power primer. *Psychological Bulletin, 112,* 155-159. doi: 10.1037/0033-2909.112.1.155

Cystic Fibrosis Foundation (2008). Patient registry. Annual Data Report 2008. Retrieved from http://www.cff.org/UploadedFiles/research/ClinicalResearch/2008-Patient-Registry-Report.pdf

Davies, J.C., & Alton, E.W.F.W (2009). Monitoring respiratory disease severity in cystic fibrosis. *Respiratory Care, 54,* 606-617.

de Abreu e Silva, F.A., & Dodge, J.A. (1996). Guidelines for the diagnosis and treatment of cystic fibrosis. WHO Human Genetics Programme and the International Cystic Fibrosis Association.

Emanuel, E.J., Wendler, D., & Grady, C. (2000). What makes clinical research ethical? *Journal of the American Medical Association, 283,* 2701-2711. Retrieved from www.jama.com

Ernst, M.M., Johnson, M.C., & Stark L.J. (2010). Developmental and Psychological issues in cystic fibrosis. *Child & Adolescent Psychiatric Clinics of North America, 19,* 263–283. doi: 10.1016/j.chc.2010.01.004

Evers, A.W.M., Kraaimaat, F.W., Lankveld, W. van, Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavorable thinking: The Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Feinstein, A.B., Forman, E.M., Masuda, A., Cohen L.L., Herbert, J.D., Moorthy, L.N., & Goldsmith, D.P. (2011). Pain intensity, psychological inflexibility, and acceptance of pain as predictor of functioning in adolescents and young adults with juvenile idiopathic arthritis: a preliminary investigation. *Journal of Clinical Psychology in Medical Settings, 18,* 291-298. doi 10.1007/s10880-011-9243-6

Geller, D.E., & Rubin, B.K. (2009). Respiratory care and cystic fibrosis. *Respiratory Care, 54,* 796-800.

Gjengedal, E., Rustøen, T., Wahl, A.K., & Hanestadt, B.R. (2003). Growing up and living with cystic fibrosis. Everyday life and encounters with the health care and social services – A qualitative study. *Advances in Nursing Science, 26 (2),* 149-159.

Goubert, L., Vervoort, T., Cano, A., & Crombez, G. (2009). Catastrophizing about their children's pain is related to higher parent-child congruency in pain ratings : an experimental investigation. *European Journal of Pain, 13,* 196-209. doi: 10.1016/j.ejpain.2008.03.009

Hayes, S.C. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapies. *Behavior Therapy, 35,* 639-665.

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: model, processes and outcomes. *Behaviour Research and Therapy, 44,* 1-25. doi:10.1016/j.brat.2005.06.006

Hofmann, S.G., Heering, S., Sawyer, A.T., & Asnaani, A. (2009). How to handle anxiety: the effects of reappraisal, acceptance, and suppression strategies on anxious arousal. *Behaviour Research and Therapy, 47,* 389-394. doi: 10.1016/j.brat.2009.02.010

Klosky, J.L., Tyc, V.L., Tong, X., Srivastava, D.K., Kronenberg, M., de Armendi, A.J., & Merchant, T.E. (2007). Perdicting pediatric distress during radiation therapy procedures: the role of medical, psychosocial, and demographic factors. *Pediatrics, 119(5),* 1159-1166. doi: 10.1542/peds.2005-1514

Lauwerier, E., Crombez, G., Van Damme, S., Goubert, L., Vogelaers, D., & Evers, A. W. M. (2010). The construct validity of the Illness Cognition Questionnaire: the robustness of the three-factor structure across patients with chronic pain and chronic fatigue. *International Journal of Behavioral Medicine, 17(2),* 90-96. doi: 10.1007/s12529-009-9059-z

Liou, T.G., Adler, F.R., FitzSimmons, S.C., Cahill, B.C., Hibbs, J.R., & Marshall, B.C. (2001). Predictive 5-year survivorship model of cystic fibrosis. *American Journal of Epidemiology, 153,* 345-352. doi: 10.1093/aje/153.4.345

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: Acceptance and Commitment Therapy for pediatric sickle cell disease. *Journal of Pediatric Psychology, 36,* 398-408. doi:10.1093/jpepsy/jsq118

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents and young adults with chronic pain: validation of an adapted assessment

instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi:10.1016/j.ejpain.2009.05.002

McCracken, L.M. & Vowles, K.E. (2008). A prospective analysis of acceptance of pain and values-based action in patients with chronic pain. Health Psychology, 27(2), 215-220. doi: 10.1037/0278-6133.27.2.215

McCracken, L.M., Vowles, K.E., & Eccleston, C. (2004). Acceptance of chronic pain: component analysis and a revised assessment method. Pain, 107, 159-166. doi:10.1016/j.pain.2003.10.012

Miller, M.R., Hankinson, J., Brusasco, V., Burgos, F., Casaburi, R., … Wanger, J. (2005). Standardisation of spirometry. *European Respiratory Journal, 26,* 319-338. doi: 10.1183/09031936.05.00034805

National Heart Lung and Blood Institute (2009). Cystic fibrosis. Retrieved December 1, 2010 from http://www.nhlbi.nih.gov/health/dci/Diseases/cf/cf_what.html

O'Sullivan, B.P., & Freedman, S.D. (2009). Cystic fibrosis. The *Lancet, 373,* 1891-1904. Retrieved from www.thelancet.com

Ratjen, F., & Döring,  G. (2003). Cystic fibrosis. *The Lancet, 361,* 681-689. Retrieved from www.thelancet.com

Rosenstein, B.J., & Zeitlin, P.L. (1998). Cystic fibrosis. *The Lancet, 351,* 277-282. Retrieved from www.thelancet.com

Rudolph, K.D., Dennig, M.D., & Weisz, J.R. (1995). Determinants and consequences of children's coping in the medical setting: conceptualization, review, and critique. *Psychological Bulletin, 118(3),* 328-357. doi: 10.1037//0033-2909.118.3.328

Schechter, N.L., Zempsky, W.T., Cohen, L.L., McGrath, P.J., McMurtry, C.M., & Bright, N.S. (2007). Pain reductions during pediatric immunizations: Evidence-based review and recommendations. *Pediatrics, 119,* 1184-1198. doi: 10.1542/peds.2006-1107

Shallcross, A., Troy, A.S., Boland, M., & Mauss, I.B. (2010). Let it be: Accepting negative emotional experiences predicts decreased negative affect and depressive symptoms. *Behaviour Research and Therapy, 48,* 921-929. doi: 10.1016/j.brat.2010.05.025

Tucker, C.L., Slifer, K.J., & Dahlquist, L.M. (2001). Reliability and validity of the brief behavioural distress scale: a measure of children's distress during invasive medical procedures. *Journal of Pediatric Psychology, 26(8),* 513-523. doi: 10.1093/jpepsy/26.8.513

Vowles, K.E., McCracken, L.M., & Eccleston, C. (2008). Patient functioning and catastrophizing in chronic pain: the mediating effects of acceptance. Health Psychology, 27(2), S136-S143. doi: 10.1037/0278-6133.27.2(Suppl.).S136

Ward, C.M., Brinkman, T., Slifer, K.J., & Paranjape, S.M. (2010). Using behavioral interventions to assist with routine procedures in children with cystic fibrosis. *Journal of Cystic Fibrosis, 9,* 150-153. doi:10.1016/j.jcf.2009.11.008

Wicksell, R.K., Dahl, J., Magnusson, B., & Olsson, G.L. (2005). Using acceptance and commitment therapy in the rehabilitation of an adolescent female with chronic pain: a case example. *Cognitive and Behavioral Practice, 12,* 415-423. doi: 10.1016/S1077-7229(05)80069-0

Wicksell, R.K., Melin, L., Lekander, M., & Olsson, G.L. (2009). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – a randomized controlled trial. *Pain, 141,* 248-257. doi:10.1016/j.pain.2008.11.006

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi:10.1016/j.pain.2011.09.003

# CHAPTER 4

## GOAL SETTING AND STRIVING IN ADOLESCENTS AND YOUNG ADULTS WITH CHRONIC ILLNESS: A COMPARISON WITH HEALTHY PEERS[1]

**ABSTRACT**

*Objective:* Research suggests that chronic illness impacts goal setting and striving in adolescents and youth. It is yet largely unknown whether adolescents and young adults with chronic illness actually differ from healthy peers on goal setting and striving. Therefore, this study (a) compared the content of health goals, (b) compared the importance, success, and difficulty of health- and non-health-related goals, and (c) explored differences in the experience of interference of non-health goals by health goals between adolescents and young adults with cystic fibrosis or diabetes and their healthy peers.

*Method:* Sixty adolescents and young adults (14-22 years) with cystic fibrosis or diabetes, and 60 healthy peers (matched on age and gender) participated. Goals in five important domains of life (i.e., school/work, relationships, leisure time, personal development, and health/body) were identified using an open goal-elicitation procedure, and rated on importance, success, difficulty, and interference by health goals.

*Results:* Adolescents and young adults with cystic fibrosis or diabetes exclusively reported goals related to illness as most important health goals. Adolescents and young adults with chronic illness and healthy peers did not differ on the reported importance, success, and difficulty of their non-health goals, except for the difficulty in attaining their relationship goal, which was perceived as less difficult in adolescents and young adults with chronic illness. Adolescents and young adults with chronic illness rated health goals as more important and more difficult to achieve than healthy peers. Surprisingly,

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chronically ill youth reported to be more successful in attaining their health goal. Also, both groups did not differ on the amount of experienced interference of non-health goals by health goals.

*Conclusion:* This study shows that goal setting and striving is largely similar in adolescents and young adults with chronic illness and their healthy peers, except for some aspects of their health goals. This suggests that our sample of adolescents and young adults with cystic fibrosis or diabetes is well-adapted to their situation, is able to integrate their illness-related goals in daily life, and feels confident in striving for their health goals.

**INTRODUCTION**

Over the past decades, the prevalence of adolescents and young adults with chronic illness has significantly increased. This increase is attributable to a rise in incidence, but also to a better survival because of substantial improvements in medical care (Halfon & Newacheck, 2010; Yeo & Sawyer, 2005). As such, there is a need to understand the challenges that adolescents and young adults with chronic illness face and how to manage these challenges (Yeo & Sawyer, 2005; World Health Organization, 2012).

Two chronic illnesses showing an upward trend are cystic fibrosis (CF; due to increased rates of survival into adulthood) and diabetes mellitus (due to a growing incidence) (Cystic Fibrosis Foundation, 2011; World Health Organization, 2012). Both are complex, invalidating, and life-shortening illnesses that require daily treatment and close medical supervision (Ward, 2010; World Health Organization, 2012). This deteriorating health and demanding treatment may impact the goal setting and striving of affected individuals. Goals are internal representations of future states one tries to attain, maintain, or avoid. These stimulate behaviour and development, thereby providing meaning and direction to life (Austin & Vancouver, 1996; Carver & Scheier, 1998).

Adolescence is a critical period for goal setting and striving. Adolescents and young adults need to achieve important developmental goals to pass on to adulthood and start to set and strive for their goals more autonomously (see Massey, Gebhardt, & Garnefski, 2008 for a review; Schwartz & Drotar, 2006, 2009). In individuals with a chronic illness, adolescence is also a period wherein substantial changes in illness may occur and full awareness of the impact of illness on one's life goals may arise. Moreover, these adolescents and young adults start to establish stable health-related goals, and are more and more able to consider the costs and benefits of these health-related goals (Badlan, 2006; Holmbeck, 2002; Schwartz & Drotar, 2006, 2009). Therefore, adolescents and young adults with chronic illness may be particularly prone to disturbances in goal setting and striving (Badlan, 2006; Maes & Karoly; 2005; Schwartz & Drotar, 2006, 2009). Investigating how adolescents and young adults with chronic illness set and strive for goals in comparison with healthy peers is a key aspect to understand how they can attain and maintain well-being and a good quality of life (Schwartz & Drotar, 2006, 2009).

Research in several chronic conditions has shown that chronic illness may affect goal setting and striving in at least three ways. First, chronic illness may lead to the

setting of health goals specifically related to illness (e.g., 'taking medicines', 'adhering to treatment'). Second, chronic illness may lead to a reprioritization and adaptation of goals so that illness-related and other life goals can be reconciled (e.g., 'putting leisure time goals on hold in favor of treatment goals', 'being a good student instead of the best student'). Third, chronic illness may lead to changes and/or hindrance in goal striving itself (e.g., having difficulties to strive for goals because of physical complaints or limitations, showing less commitment to goals) (see Schwartz & Drotar, 2006 for a review).

Despite growing attention for goal regulation in paediatric chronic illness (e.g., Helgeson & Takeda, 2009; Schwartz & Drotar, 2006, 2009; Schwartz & Parisi, in press; Seiffge-Krenke, 1998), there are to the best of our knowledge, only two studies that compared goal setting and striving in adolescents and young adults with diabetes with that in healthy peers (Helgeson & Takeda, 2009; Seiffge-Krenke,1998), and no studies that make this comparison for adolescents and young adults with CF. These studies found that adolescents and young adults with diabetes did not differ in the number of identified life goals from healthy peers. It was also found that adolescents and young adults with diabetes mostly shared the same developmental goals but identified less self-improvement and more appearance goals compared with their healthy peers. Furthermore, adolescents and young adults with diabetes perceived delays in some of their developmental goals (i.e., physical maturity and individual lifestyle) compared with healthy adolescents and young adults (Helgeson & Takeda, 2009; Seiffge-Krenke, 1998). These findings are not in line with recent research suggesting that adolescents with cancer identify fewer life goals, are less likely to identify interpersonal or leisure goals, and rate their goals as more achievable as compared with healthy peers (Schwartz & Parisi, in press). These studies, however, did not focus on the specific aspects of goal setting and striving that are assumed to be affected by chronic illness (i.e., new health goals, reprioritization and adaptation of goals, and changes and/or hindrance in goal striving). Consequently, objective of the current study was to compare adolescents and young adults with CF and diabetes with healthy peers on these aspects. Specifically, as chronic illness may lead to the setting of new goals related to illness, a first aim of this study was to compare the content of health goals between adolescents and young adults with CF/diabetes and their healthy peers. Second, as chronic illness may force to reprioritize and adapt goals, this study aimed to compare the importance of health- and non-health-related goals between these adolescents and young adults. A third aim of this study,

bearing in mind the potential changes and/or hindrance in goal striving due to chronic illness, was to compare the goal striving of these adolescents and young adults in terms of success and difficulty. A final aim, given the potential setting of new goals and reprioritization and adoption of goals, was to explore the experience of interference of non-health goals by health goals.

Adolescents and young adults were free to report as many goals as they wanted in five domains of life: school/work, relationships, leisure time, personal development, and health/body (Massey, Gebhardt, & Garnefski, 2008). It was hypothesized that the health goals that adolescents and young adults with CF or diabetes identified as most important relate to their illness. Also, it was expected that youth with illness would rate their health goals as more important compared with healthy peers. Finally, it was expected that adolescents and young adults with CF and diabetes report to be less successful and experience more difficulties in attaining their health and non-health goals as compared with healthy peers.

## METHOD

This study is part of the Goals, Acceptance, and Well-being in Chronic Illness (GAWCI) study, performed between September 2010 and April 2012 in Flanders, Belgium. Participants for the GAWCI study were adolescents and young adults with CF or diabetes and matched (age and gender) healthy peers. To recruit the adolescents and young adults with CF, four University Hospitals were contacted for participation. The University Hospitals of Ghent and Leuven agreed to take part in the study. To recruit the adolescents and young adults with diabetes, the University Hospital of Ghent was contacted and agreed to participate. The healthy peers were recruited from a sample of school children who participated between 2008 and 2011 in one of two questionnaire studies, see Caes, Vervoort, Eccleston, & Goubert, 2012 and Vervoort, Huguet, Verhoeven, & Goubert, 2011, and had given consent to be re-contacted for other studies.

### Participants

Inclusion criteria for participation were: 14-22 years of age, comprehension of the Dutch language, and no developmental disorder. Furthermore, adolescents and young adults with CF who had/were awaiting lung transplantation were excluded because of the exceptional status of this situation (e.g., very high level of symptoms, being terminally ill, intertwinement of end of life issues and hope of transplantation) (Bourke et al., 2009). One hundred and thirty adolescents and young adults, of whom 74 had CF and 56 had

diabetes, met the inclusion criteria. All eligible participants were invited by letter and/or recruited during routine clinic visits by the psychologist from the respective hospital (time period of recruitment: September 2010-April 2011). Subsequently, adolescents and young adults who agreed to participate (*N* = 82) where phoned by a research assistant and a home visit was planned. Finally, 60 adolescents and young adults (CF = 35, diabetes = 25; 26 males, 34 females; response rate = 46.15%) with CF or diabetes, and 60 healthy peers, matched on age and gender, took part in the GAWCI study. To obtain the healthy peers, 121 eligible healthy adolescents and young adults were contacted by phone (response rate = 49.59%). The main reasons for not participating were a lack of time and interest. All participants were of Caucasian origin. Mean age was 17.43 years (*SD* = 2.67) for the adolescents and young adults with CF or diabetes, and 17.80 years (*SD* = 2.71) for healthy peers. *T*-tests revealed that mean age of adolescents and young adults with chronic illness was comparable to mean age of the healthy peers (*t*(118) = -.77, *p* = .445). Of the non- participants with CF or diabetes, 55.71% was diagnosed with CF and 44.29% with diabetes. Of the healthy non-participants 48.33% were male and 51.67% female. Because of confidentiality, no further data about the characteristics of the non-participants were available.

**Procedure**

A research assistant visited the participants at home. During this visit, written parent consent (adolescents and young adults < 18 years) and/or adolescent assent (adolescents and young adults < 18 years)/consent (adolescents and young adults > 18 years) were obtained and a set of questionnaires was completed under supervision of a research assistant. Of this set, only the questionnaires concerning goals were included in the present study. The GAWCI study was approved by the ethical committees of the University Hospitals of Ghent and Leuven, and was carried out in accordance with universal ethical principles (Emanuel, Wendler, & Grady, 2000).

**Measures**

*Socio-demographic information* (age, gender, time since diagnosis, type of diagnosis) was verbally inquired before administration of the questionnaires.

*Goals* were explored using an open goal-elicitation procedure. Based upon earlier research (see Massey, Gebhardt, & Garnefski, 2008), participants were asked to report as many goals as they wanted regarding the following five domains: school/work, relationships, leisure time, personal development (yourself), and health/body. In line with Massey and colleagues (2009), goals were elicited by means of the following instruction:

'Some things you find important in life, other things you find less important. People differ in this respect: what one considers important and consequently has as a goal, can be less important for another. Everyone has his or her own goals, and that makes every person unique and special. Some goals you like to attain, other goals matter less. We would like to ask you some questions concerning the things you consider important in life. In other words: what are your most important goals? It is important to know that : (a) one can have different goals in different domains at the same time, for example in the domain of school, relationships, work, yourself, leisure time, or health, (b) goals have to do with either wanting to do something or exactly not wanting to do it. We are interested in the goals that you are currently pursuing, and that will still be important for you within a few months.'. After the elicitation procedure, all participants were asked to select their most important goal per domain. In line with the Personal Projects Analysis from Little (1983), these five goals were then rated on importance ("How important is this goal to you?" 0 = not at all, 10 = very much), success ("How successful are you in attaining this goal?" 0 = not at all, 10 = very much), and difficulty ("How difficult is it to attain this goal?" 0 = not at all, 10 = very much). This procedure was pilot tested on comprehensibility and relevance in eight healthy adolescents and young adults. As this pilot test was successful, no modifications to the original procedure were made.

*Goal interference* was measured with an adapted version of Little's Personal Projects Cross-Impact Matrix (1983). New in this version was that the negative influence of one goal on another (i.e., 'goal interference') was considered independent of its positive influence. All participants were requested to rate to what extent their health/body goal had a negative influence on their most important school/work, relationships, free time, and personal development goal. All items were rated on an 11-point Likert scale (0 = no negative influence, 10 = strong negative influence), with higher scores indicating a more negative impact. A mean score of these four items was calculated. The adapted matrix was successfully tested on comprehensibility and relevance in a pilot study with eight healthy adolescents and young adults.

**Data analysis**

All statistical analyses were performed using SPSS 20.0. First, some preliminary analyses were performed. Tests of normality (i.e., Kolmogorov-Smirnov test) were performed for all continuous variables. Descriptive statistics (means and *SD*'s) were computed for all variables of interest. To test for differences between adolescents and

young adults with CF or diabetes and their healthy peers regarding their goals, a comparison of means between two independent groups was performed.


**RESULTS**

Tests of normality (i.e., Kolmogorov-Smirnov tests) revealed that the scores for the importance, success, and difficulty of the reported goals, and the scores for interference of life goals by health goals were non-normally distributed. The scores on goal importance, success, and difficulty were skewed to the left, whereas scores for goal interference were skewed to the right. Therefore, the Mann-Whitney U Test was performed to identify potential differences between the goals of adolescents and young adults with CF/diabetes and those of their healthy peers.

**Descriptive statistics**

Means and *SD*'s of the variables of interest are reported in Table 1 for each group.

**Health goal content**

Health goals identified as most important by the adolescents and young adults with CF or diabetes related to illness. In adolescents and young adults with CF, reported health goals were 'stay healthy/stable' ($N = 7$), '(stay) engage(d) in treatment' ($N = 7$), 'take good care of myself to prevent deterioration' ($N = 4$), 'gain weight/eat well' ($N = 4$), (do more sports to) increase my lung functioning' ($N = 4$), '(not forget to) take my medicines (on time)' ($N = 2$), 'obtain a better/preserve a good condition' ($N = 2$), 'perform better in physiotherapy' ($N = 2$), 'avoid infections' ($N = 1$), 'avoid hospitalization' ($N = 1$), 'take my aerosol better' ($N = 1$). Health goals reported by adolescents and young adults with diabetes were 'get lower/better blood sugar levels' ($N = 6$), 'eat healthy' ($N = 4$), 'do more sports' ($N = 4$), 'lose weight' ($N = 3$), 'have a healthy life style' ($N = 2$), 'adhere to my diet' ($N = 1$), 'get control over my diabetes' ($N = 1$), 'become less ill' ($N = 1$), 'stay healthy' ($N = 1$), 'get rid of my diabetes' ($N = 1$), 'cope with my diabetes' ($N = 1$). Examples of health goals frequently reported by healthy peers were 'exercise more' and 'eat healthy'.

Table 1

*Means (M) and standard deviations (SD) of the importance, success, and difficulty of goals and experienced goal interference of non-health goals by health goals*

|  | $M_{CF,diabetes}$ | $SD_{CF,diabetes}$ | $M_{healthy}$ | $SD_{healthy}$ |
|---|---|---|---|---|
| *School/work goal* |  |  |  |  |
| 1. Importance | 8.60 | 1.53 | 8.83 | 1.11 |
| 2. Success | 6.52 | 1.96 | 6.67 | 1.89 |
| 3. Difficulty | 6.17 | 1.98 | 5.92 | 2.32 |
| *Relationships goal* |  |  |  |  |
| 1. Importance | 8.63 | 1.48 | 8.35 | 1.27 |
| 2. Success | 6.97 | 2.53 | 6.25 | 2.47 |
| 3. Difficulty | 4.35 | 2.71 | 5.53 | 2.61 |
| *Leisure time goal* |  |  |  |  |
| 1. Importance | 7.88 | 1.87 | 7.67 | 1.13 |
| 2. Success | 6.77 | 2.03 | 6.33 | 2.34 |
| 3. Difficulty | 4.85 | 2.56 | 4.83 | 2.56 |
| *Yourself goal* |  |  |  |  |
| 1. Importance | 7.70 | 1.96 | 7.90 | 1.34 |
| 2. Success | 5.73 | 2.59 | 5.33 | 2.21 |
| 3. Difficulty | 6.27 | 2.67 | 6.45 | 2.30 |
| *Health goal* |  |  |  |  |
| 1. Importance | 9.07 | 1.36 | 7.52 | 1.35 |
| 2. Success | 6.55 | 1.64 | 5.58 | 2.11 |
| 3.Diffculty | 6.90 | 1.97 | 5.60 | 2.44 |
| *Goal interference* | 2.01 | .26 | 1.93 | .25 |

*$p < .05$; **$p < .01$

## Group differences in goal importance, success, difficulty, and interference

There were no significant differences between adolescents and young adults with CF or diabetes and their healthy peers on the importance of their goals regarding school/work ($U = 1673.500$, $p = .490$), relationships ($U = 1515$, $p = .124$), leisure time ($U = 1485$, $p = .090$), and personal development ($U = 1774$, $p = .889$). However, both groups did differ on the importance of their health goal ($U = 680.500$, $p = .000$), with adolescents and young adults with CF or diabetes rating their health goal as more important. Also, adolescents and young adults with C or diabetes and their healthy peers did not differ significantly on the reported success of attaining their goals regarding school/work ($U = 1672$, $p = .493$), relationships ($U = 1446$, $p = .060$), leisure time ($U = 1639$, $p = .391$), and personal development ($U = 1603$, $p = .296$), but they did differ on the success of their health goal ($U = 1309$, $p = .009$). Interestingly, adolescents and young adults with CF or

diabetes rated themselves as more successful in attaining their health goal. Furthermore, both groups did not differ on the level of difficulty in attaining their school/work goal ($U$ = 1718, $p$ = .663), leisure time goal ($U$ = 1778,500, $p$ = .909), and personal development goal ($U$ = 1794, $p$ = .975). But they did differ on the level of reported difficulty in attaining their relationship goal ($U$ = 1339,500, $p$ = .015) and health goal ($U$ = 1228, $p$ = .002). Youth with chronic illness rated their relationship goal as less difficult and their health goal as more difficult to attain. Finally, both groups reported equal levels of interference of life goals by their most important health goal ($U$ = 1786, $p$ = .941).

## DISCUSSION

Aim of the current study was to compare goal setting and striving in adolescents and young adults with CF or diabetes with healthy peers. We explicitly focused on differences between adolescents and young adults with CF or diabetes and healthy peers in those aspects of goal setting and striving that are assumed to be affected by chronic illness. In other words, the current study is the first to compare the content of health goals between adolescents and young adults with CF or diabetes and their healthy peers, to compare the importance of their (non-)health goals, and the success, and difficulty in attaining these goals between these adolescents and young adults, and to compare the amount of experienced interference between their health and non-health goals.

In line with our expectations, it was found that adolescents and young adults with CF or diabetes, in contrast to their healthy peers, exclusively reported goals relating to their illness as their most important health goals. Moreover, chronically ill youth rated their health goals as more important compared with the health goals of their healthy peers, but rated their non-health goals as equally important. Furthermore, adolescents and young adults with CF or diabetes rated their health goals as more difficult to attain in comparison with their healthy peers, but they did not differ on the experienced difficulty in attaining their school/work, relationships, leisure time, and personal development goals), except for the level of difficulty of their relationship goal, which was reported as less difficult to attain for adolescents and young adults with chronic illness. Intriguingly, adolescents and young adults with CF or diabetes rated themselves as more successful in attaining their health-goal as compared with healthy peers but did not differ on the amount of success in attaining their school/work, relationships, leisure time, and personal development goals. Finally, there were no differences between ill and healthy youth in experienced interference of non-health goals by health goals.

Our results suggest that goal setting and striving is largely similar in adolescents and young adults with and without CF or diabetes, except for their goals regarding health. The finding that adolescents and young adults with CF or diabetes identified goals related to their illness as their most important health goals, is in line with previous research stating that chronic illness forces to set new goals in response to this illness (e.g., 'taking medicines', 'adhering to treatment'; see Schwartz & Drotar, 2006 for a review). Also, adolescents and young adults with chronic illness rated their health goals as more important compared with their healthy peers. The finding that both groups rated their non-health goals equally in terms of importance may indicate that adolescents and young adults with CF or diabetes give the same priority to these goals as their healthy peers. This observation suggests that adolescents and young adults with CF and diabetes are able to adapt and incorporate their goals so that both health and non-health goals can be reconciled (see Schwartz & Drotar, 2006 for a review). Specific strategies that may be used to tune different goals to each other without scaling back their priority, are the use of compatible strategies to attain different goals (e.g., 'join a running team' may be effective for both 'obtain a better lung functioning' and 'meet new people'), the identification of possible instrumental relations between goals (e.g., 'take responsibility for treatment' may be a step towards 'become independent'), and the optimization of the use of limited resources (e.g., time, energy; e.g., 'doing aerosol treatment while studying') (Riediger & Freund, 2004).

We also found that adolescents and young adults with CF or diabetes reported more success in attaining their health goals, but rated their health goals also as more difficult to attain compared with their healthy peers. Only the latter finding, but not the first, seems to fit with previous research indicating that chronic illness inevitably leads to changes and/or hindrance in goal striving itself (e.g., having difficulties to strive for goals because of physical complaints or limitations, showing less commitment to goals). The latter finding, however, can also be explained by the fact that goals relating to illness are more complex and demanding than non-illness-related health goals (Badlan, 2006; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2006, 2009; Seiffge-Krenke, 2001). Possible explanation for the seeming contradiction between these findings concerning success and difficulty may be that chronically ill adolescents and young adults may have a high sense of self-efficacy concerning their health goals, have a stronger commitment to their health goals, or possess certain self-regulatory skills (i.e., planning, self-monitoring), whereby more difficulty does not necessarily translates into less success (Maes & Karoly,

2005). The current study also found that adolescents and young adults with CF or diabetes and their healthy peers rated themselves as equally successful in attaining their non-health goals and rated their non-health goals (except for their relationships goals) as equally difficult to attain. Furthermore, both groups reported equal levels of interference of non-health goals by illness goals. These findings are not in line with previous research suggesting that chronic illness may cause hindrance in goal striving (see Schwartz & Drotar, 2006 for a review). Rather it suggests that the chronically ill adolescents and young adults in this study are able to balance the demands of chronic illness with their regular life goals, without disengaging from or scaling back on these goals (cfr. equal ratings on goal importance). Ways to obtain such balance may be, on top of the strategies mentioned above, the adoption of more realistic goals or the adjustment of goals to situational constraints (e.g., obtain a college instead of a university degree) (Brandstätter & Renner, 1990; Schwartz & Drotar, 2009). Future research should investigate which strategies adolescents and young adults with chronic illness exactly use to maintain and integrate their health and non-health goals.

Adolescents and young adults with and without CF or diabetes seem to share more similarities than differences regarding their non-health goals. Nonetheless, it was clear that goal setting and striving in adolescents and young adults with CF and diabetes was affected by illness given the identification and prioritization of goals related to their illness. However, despite this focus on illness-related goals these adolescents and young adults were overall equally interested, engaged, and effective in other life goals as compared with their peers, except for the difficulty in attaining their relationship goals. This seems to suggest that the adolescents and young adults with CF and diabetes from the current study were well-adapted to their situation, had developed effective strategies to integrate their illness-related goals in daily life, and seemed to feel confident in striving for their health goals.

Some limitations of the current study should be considered when interpreting the results. First, only 60 of the 130 eligible adolescents and young adults with CF and diabetes participated. It is possible that participating adolescents and young adults have characteristics that do not generalize to non-participating adolescents and young adults (e.g., well-adapted, experiencing low goal interference). Second, the lack of evidence for differences in goal setting and striving does not suggest a generalizable absence of differences as it depends on statistical power. Third, this study only included adolescents and young adults with CF or diabetes. Therefore, it is not known whether the current

results also apply for adolescents and young adults with other chronic conditions. Future research should further investigate this by including several chronic conditions. Fourth, the current study did not focus on how adolescents and young adults with CF or diabetes manage to incorporate or reconcile their health and non-health goals. Further research is needed to understand what strategies these adolescents and young adults use to manage this. Additionally, longitudinal designs may shed light on how adolescents and young adults with chronic illness, from the time of diagnosis on, set, pursuit, adjust, attain, and balance their health and non-health goals.

In clinical practice it is necessary to carefully monitor the goal setting and striving of adolescents and young adults with CF and diabetes and to intervene when difficulties occur. Such difficulties could include the experience of life goal interference by health goals, unnecessary disengagement from valued and essential life goals, unbalanced health and non-health goals, etc. Because goals stimulate behaviour and development, relate to well-being and may affect adherence, the timely notice of such difficulties is of importance (Austin & Vancouver, 1996; Carver & Scheier, 1998; Schwartz & Drotar, 2006). Self-regulation interventions incorporating the strategies mentioned above may prove useful to solve these difficulties.

## REFERENCES

Austin, J. T., & Vancouver, J. B. (1996). Goal constructs in psychology: Structure, process, and content. *Psychological Bulletin, 120,* 338–375. doi:10.1037/0033-2909.120.3.338

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14,* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Bourke, S. J., Doe, S. J., Gascoigne, A. D., Heslop, K., Fields, M., Reynolds, D., & Mannix, K. (2009). An integrated model of provision of palliative care to patients

with    cystic    fibrosis.    *Palliative    Medicine,    23,*    512–517.    doi:
10.1177/0269216309106312

Brandstätter, J., & Renner, G. (1990). Tenacious goal pursuit and flexible goal
adjustment: explication and age-related analysis of assimilative and accommodative
strategies of coping. *Psychology and Aging, 5,* 58-67.

Caes, L., Vervoort, T., Eccleston C., & Goubert, L. (2012). Parents who catastrophize
about their child's pain prioritize attempts to control pain. *Pain, 153,* 1695-1701.

Carver, C. S., & Scheier, M. F. (1998). On the Self-Regulation of Behavior. New York,
NY: Cambridge University Press.

Cystic Fibrosis Foundation (2011). Annual Report. Adding tomorrows. Retrieved from
http://www.cff.org/UploadedFiles/aboutCFFoundation/AnnualReport/2011-Annual-
Report.pdf

Emanuel, E.J., Wendler, D., Grady, C. (2000). What makes clinical research ethical?
*JAMA, 283,* 2701-2711. Retrieved from www.jama.com

Glasscoe, C. A., & Quittner, A. L. Psychological interventions for people with cystic
fibrosis and their families. Cochrane Database of Systematic Reviews 2008, Issue 3.
Art. No.: CD003148. doi: 10.1002/14651858.CD003148.pub2.

Halfon, N., & Newacheck, P.W. (2010). Evolving notions of childhood chronic illness.
*JAMA, 303,* 665-666. Retrieved from jama.ama-assn.org

Helgeson, V. S., & Takeda, A. (2009). Brief report: Nature and implications of personal
projects among adolescents with and without diabetes. *Journal of Pediatric
Psychology, 34,* 1019–1024. doi:10.1093/jpepsy/jsp024

Holmbeck, G. (2002). A developmental perspective on adolescent health and illness: an
introduction to the special issues. *Journal of Pediatric Psychology, 27(5),* 409-415.

Little,    B.    (1983).    Personal    Projects    Analysis.    Retrieved    from
http://www.brianrlittle.com/Topics/research/assessment-tools/

Maes, S., & Karoly, P. (2005). Self-regulation assessment and intervention in physical
health and illness: A review. *Applied Psychology, 54,* 267–299. doi:10.1111/j.1464-
0597.2005.00210.x

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and
pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,*
421-460. doi: 10.1016/j.dr.2008.03.002

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2009). Self-generated goals and goal process appraisals: Relationships with sociodemographic factors and well-being. *Journal of Adolescence, 32,* 501-518. doi: 10.1016/j.adolescence.2008.07.003

Riediger, M. & Freund, A.M. (2004). Interference and facilitation among personal goals: differential associations with subjective well-being and persistent goal pursuit. *Personality and Social Psychology Bulletin, 30,* 1511-1523. doi: 10.1177/0146167204271184

Schwartz, L. A., & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: A review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 390–405. doi:10.1007/s10880-006-9041-8

Schwartz, L. A., & Drotar, D. (2009). Health-related hindrance of personal coal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 39,* 954–965. doi:10.1093/jpepsy/jsn144

Seiffge-Krenke, I. (1998). Chronic disease and perceived developmental progression in adolescence. *Developmental Psychology, 34,* 1073–1084. doi:10.1037/0012-1649.34.5.1073

Vervoort, T., Huguet, A., Verhoeven, K., & Goubert, L. (2011). Mothers' and fathers' responses to their child's pain catastrophizing and disability. *Pain, 152,* 786-793. doi: 10.1016/j.pain.2010.12.010

Ward, C.M., Brinkman, T., Slifer, K.J., & Paranjape, S.M. (2010). Using behavioral interventions to assist with routine procedures in children with cystic fibrosis. *Journal of Cystic Fibrosis, 9,* 150-153. doi: 10.1016/j.jcf.2009.11.008

World Health Organization (2012). What are the risks of diabetes in children? Retrieved from http://www.who.int/features/qa/65/en/index.html

Yeo, M., & Sawyer, S. (2005). ABC of adolescence. Chronic illness and disability. *British Medical Journal, 330,* 721-723. doi: 10.1136/bmj.330.7493.721

# CHAPTER 5

## ACCEPTANCE, WELL-BEING, AND GOALS IN ADOLESCENTS WITH CHRONIC ILLNESS: A DAILY PROCESS ANALYSIS [1]

**ABSTRACT**

*Objective:* The main aim of this study was to investigate the relationship between acceptance and well-being in adolescents with chronic illness from a daily process perspective. Furthermore, we explored the role of daily experienced interference and facilitation of life goals by treatment goals as mediating mechanisms.

*Method:* Thirty-eight adolescents with cystic fibrosis or diabetes completed questionnaires assessing acceptance, negative life events and goal-related self-efficacy. Furthermore, an online diary assessing daily mood, daily experienced interference and facilitation of life goals by treatment goals was completed during three consecutive weeks.

*Results:* Acceptance of illness was positively related to daily well-being, but unrelated to daily goal interference and facilitation. Furthermore, daily goal interference and facilitation were unrelated to same and next day well-being.

*Conclusion:* This study suggests that acceptance of illness plays an important role in the daily mood of adolescents with cystic fibrosis or diabetes. This relationship, however, was not mediated by daily experienced interference and facilitation of life goals by treatment goals. Further research is needed to determine whether interventions promoting acceptance are beneficial for adolescents with cystic fibrosis and diabetes.

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**INTRODUCTION**

Previous research has indicated that chronic illness may seriously impact upon the well-being of adolescents. More specifically, growing up with chronic illness is a risk factor for several problems such as depressive symptoms, behaviour problems, and impairment in academic, physical, and social functioning (see Pinquart & Shen, 2011; Pinquart & Teubert, 2012). It has been suggested that the way of evaluating illness plays an important role in well-being (Evers et al., 2001; Maes & Karoly, 2005). A specific type of evaluation that may be relevant is 'acceptance'. Acceptance can be defined as 'recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences' (Evers et al., 2001, p. 1027). Questionnaire studies have shown that acceptance may have a protective role in the well-being of adolescents with chronic illnesses such as chronic pain (e.g., McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011), juvenile arthritis (Feinstein et al., 2011), sickle cell disease (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011), and cystic fibrosis (e.g., Casier et al., 2011). Specifically, this research has shown that higher acceptance of chronic illness is related to positive outcomes such as less anxiety, less depression, less disability, and better emotional, social, and physical functioning. The present study will contribute to this literature by examining whether acceptance of illness is also related to (fluctuations in) daily well-being (i.e., positive/negative mood).

As yet, no research has addressed underlying processes. As chronic illness creates the need to pursue additional treatment goals to keep an optimal level of physical functioning and well-being, the experience of interference and/or facilitation of life goals (e.g., academic performance) by these treatment goals may be potential candidates. Goal interference occurs when striving for one goal (e.g., adhering to treatment) decreases the chance of attaining another goal (e.g., becoming independent) and can arise from limited resources for goal striving (e.g., time, energy) and/or from incompatible goal striving strategies (Little, 1983a; Riediger & Freund, 2004). Goal facilitation occurs when striving for one goal increases the chance of attaining another goal (e.g., the goal 'doing treatment' may facilitate the goal 'improving the relationship with my parents' as it may lead to less tension between adolescents and their parents). Reasons for investigating goal interference and facilitation as possible underlying mechanisms in the relationship between acceptance and well-being are twofold. First, acceptance is assumed as having a key role in the engagement in important life goals, despite being ill (Evers et al., 2001;

Hayes, Luoma, Bond, Masuda, Lillis, 2006; Maes & Karoly, 2005; Risdon, Eccleston, Crombez, & McCracken, 2003). Consequently, acceptance may also relate to interactions between treatment and life goals, and specifically, to the extent to which these goals interfere with and/or facilitate each other. Second, goal processes have increasingly been suggested as impacting well-being in the context of chronic illness (De Ridder & Kuijer, 2007; Gebhardt, 2008). Indeed, previous research in healthy individuals has shown that goal interference is related to lower levels of well-being, whereas goal facilitation is mostly unrelated, but sometimes positively related to well-being (see Riediger, 2007 for an overview). If this is also the case in individuals with chronic illness is not yet known and has to be empirically investigated. Insight in how acceptance impacts well-being, and the role of intergoal interference/facilitation is particularly relevant in adolescence since achieving important life goals in the transition to adulthood may be more difficult for adolescents with chronic illness than for healthy peers (Badlan, 2006; Schwartz & Drotar, 2006).

The primary aim of the present study was to examine the relationship between acceptance and well-being (i.e., daily mood) in adolescents with chronic illness on a daily basis. The secondary aim was to investigate whether acceptance was related to daily goal interference and facilitation, and whether daily goal interference/facilitation was related to well-being. Thirty-eight adolescents with chronic illness reported on daily well-being (i.e., positive/negative mood), daily experienced interference and facilitation of life goals by treatment goals. A daily process perspective allows investigating the experiences of adolescents in their natural context, capturing experiences closely to their occurrence, and reducing the chance of recall bias (Bolger, Davis, & Rafaeli, 2003). It was hypothesized that higher levels of acceptance are related to higher daily positive mood, lower daily negative mood, and to less experienced interference in daily life goals. Furthermore, we expected higher experienced interference in daily life goals to be related to lower (same and next day) positive mood and higher (same and next day) negative mood. Finally, the relationships among acceptance, goal facilitation and daily mood were explored, as well as the mediating role of goal interference and facilitation in the relationship between acceptance and daily mood. Given the potential impact of negative life events and goal-related self-efficacy upon goal pursuit, we controlled for these variables when testing the hypotheses concerning goals.

**METHOD**

This study is part of the Goals, Acceptance, and Well-being in Chronic Illness (GAWCI) study, performed between September 2010 and April 2012 in Flanders, Belgium. Participants for the GAWCI study were adolescents with cystic fibrosis (CF) and diabetes and matched (age and gender) healthy peers. The healthy peers (recruited from a sample of school children who participated between 2008 and 2011 in one of two questionnaire studies, see Caes, Vervoort, Eccleston, & Goubert, in revision and Vervoort, Huguet, Verhoeven, & Goubert, 2011, and had given consent to be re-contacted for other studies) were included in an independent part of the GAWCI study not reported here. To recruit the adolescents with CF all four Flemish University Hospitals were contacted for participation. The University Hospitals of Ghent and Leuven agreed to take part in the study. To recruit the adolescents with diabetes, the University Hospital of Ghent was contacted and agreed to participate.

**Participants**

Inclusion criteria for individual participation were: 14-22 years of age, comprehension of the Dutch language, and no developmental disorder. Furthermore, adolescents with CF who had/were awaiting lung transplantation were excluded because of the exceptional status of this situation (e.g., very high level of symptoms, being terminally ill, intertwinement of end of life issues and hope of transplantation) (Bourke et al., 2009). One hundred and thirty adolescents, of whom 74 had CF and 56 had diabetes, met the inclusion criteria. All eligible participants were invited by letter and/or recruited during routine clinic visits by the psychologist from the respective hospital (time period of recruitment: September 2010-April 2011). Subsequently, adolescents who agreed to participate ($N = 82$) where phoned by a research assistant and a home visit was planned. Finally, 62 adolescents (27 males, 35 females; response rate = 47.69%) with CF and diabetes took part in the GAWCI study. The main reasons for not participating were a lack of time and interest. Of the 62 adolescents enrolled in the study, 58 agreed to complete the diary. In case less than 75% of the requested diaries were completed, data were excluded from further analysis (Huijnen, Verbunt, Roelofs, Goossens, & Peters, 2009), resulting in a final sample of 38 adolescents (participation rate = 29%; CF: $N = 22$, 12 males, 10 females; diabetes: $N = 16$, 6 males, 10 females). All adolescents were of Caucasian origin. Mean age was 18.63 years ($SD = 2.75$) for the adolescents with CF and 16.30 years ($SD = 1.58$) for the adolescents with diabetes. Mean time since diagnosis was 16.28 years ($SD = 5.83$) for the adolescents with CF, and 8.07 years ($SD = 4.46$) for the

adolescents with diabetes. *T*-tests revealed that, in comparison with adolescents with diabetes, adolescents with CF were significantly older ($M_c = 18.63$, $SD_c = 2.75$; $M_d = 16.30$, $SD_d = 1.58$; $t = 3.30$, $p = .00$). Of the non- participants, 56.52% was diagnosed with CF and 43.48% with diabetes. Because of confidentiality, no further data about the characteristics of the non-participants were available.

**Procedure**

First, a research assistant visited the participants at home. During this visit, written parent consent (adolescents < 18 years) and/or adolescent assent (adolescents < 18 years)/consent (adolescents > 18 years) were obtained and a set of questionnaires was completed under supervision of a research assistant. Of this set, only the questionnaires concerning goal content, acceptance of illness, negative life events, and goal-related self-efficacy were included in the present study. At the end of this home visit the research assistant explained the diary procedure. After the home visit, each participant's self-generated most important goals were entered in a personal online diary created with LimeSurvey 1.85 software. Then, each adolescent received an e-mail containing a link to their personal diary accompanied by explicit guidelines on how to complete it. In this diary, daily experienced interference and facilitation of personal goals by treatment goals were assessed during three consecutive weeks, along with daily mood and the extent to which goal pursuit was hindered by daily physical complaints. For the present study, only the diary questions on goal interference and facilitation and daily mood were used. Additional paper versions of the diary, to be used in case of computer or internet problems, were sent by regular mail. Participants were instructed to complete the diary at the end of the day. To reduce the number of missing data to a minimum every participant daily received a reminding text message (SMS) at 7.00 p.m. All participants received a financial compensation of € 30 for computer and internet use. The ethical committees of the University Hospitals of Ghent and Leuven approved the GAWCI study.

**Measures**

*Questionnaires*

*Socio-demographic information* (age, gender, time since diagnosis, type of diagnosis) was verbally inquired before administration of the questionnaires.

*Acceptance* was assessed by a subscale of the Dutch version of the Illness Cognition Questionnaire (ICQ; Evers et al., 2001). Acceptance is conceptualised as the perceived ability to live with the illness and to master its negative consequences (six items, e.g. "I can handle the problems related to my illness"). Items are rated on a 4-point

Likert scale (1 = *not at all*, 4 = *completely*). The total score for acceptance varies between 6 and 24, with higher scores indicating higher levels of acceptance. Psychometric research in samples of Dutch-speaking adults with rheumatoid arthritis, multiple sclerosis, chronic pain and chronic fatigue demonstrated the adequate reliability and validity of the ICQ (Evers et al., 2001; Lauwerier et al., 2010). Reliability of the acceptance scale in the research sample of the present study was good with a Cronbach's α of .82.

*Goals* were explored using an open goal-elicitation procedure. To ensure that several domains of life were considered during this elicitation, all adolescents were asked to report their goals for the following five domains: school/work, relationships, leisure time, yourself (personal development), and health/body. This categorization of goal domains was used as it is consistently supported by earlier goal research in adolescents (for a review see Massey, Gebhardt, & Garnefski, 2008). Goals were generated by the adolescents themselves instead of externally determined, what made it personally relevant. Adolescents were free to report as many goals as they wanted. Following Massey and colleagues (2009a), goals were elicited by means of the following instruction: 'Some things you find important in life, other things you find less important. People differ in this respect: what one considers important and consequently has as a goal, can be less important for another. Everyone has his or her own goals, and that makes every person unique and special. Some goals you like to attain, other goals matter less. We would like to ask you some questions concerning the things you consider important in life. In other words: what are your most important goals? It is important to know that : (a) one can have different goals in different domains at the same time, for example in the domain of school, relationships, work, yourself, leisure time, or health, (b) goals have to do with either wanting to do something or exactly not wanting to do it. We are interested in the goals that you are currently pursuing, and that will still be important for you within a few months.'. After the elicitation procedure, all adolescents were asked to select their most important goal per domain, except for the health domain. As the current study focused on the interrelationship of personal goals with treatment goals, the goal 'doing my treatment' was fixed as most important within this domain. This procedure was pilot tested on comprehensibility and relevance in eight healthy adolescents. As this pilot test was successful, no modifications to the original procedure were made.

*Negative life events* were measured by using a 12-item lifetime checklist for adolescents adapted from Kraaij et al. (2003). This checklist measures negative life events concerning self or significant others during the past year, such as death of someone close, parental divorce, and abuse experiences. Sum scores are computed by adding up all negative life events experienced throughout life (range 0-12).

*Goal-related self-efficacy* refers to the perception of self-efficacy in relation to being able to achieve one's goals despite possible obstacles and was measured by an adaptation of Schwarzer's General Self-Efficacy Scale (see Massey et al., 2009a). The scale consists of 10 items (e.g., "I can always manage to achieve my goals if I try hard enough") and is rated on a 4-point Likert scale (1 = *not at all true*, 4 = *exactly true*). A mean score of these 10 items was calculated (range 1-4), with higher scores indicating more goal related self-efficacy. Reliability in the present study was good with a Cronbach's α of .80.

*Health status* was subjectively rated on a 5-point Likert scale ("How would you describe your current health status?"; 1 = *bad*, 5 = *excellent*).

**Diary**

*Daily mood* was assessed with the Brief Mood Rating Scale (Diener & Emmons, 1985). This nine-item scale measures positive (four items, e.g., "happy", "joyful", "enjoyment/fun", "pleased") and negative mood (five items, e.g., "depressed/blue", "unhappy", "frustrated", "angry/hostile", "worried/anxious"). Items are rated on a 7-point Likert scale (0 = *not at all*, 6 = *extremely much*). Total scores range from 0 to 24 for positive mood and from 0 to 30 for negative mood, with higher scores indicating higher positive, respectively negative mood. The scale has been reported to have good reliability and validity properties (Diener & Emmons, 1985). Cronbach's α in the current sample was .98 for positive and .90 for negative mood.

*Goal facilitation and interference* were measured with an adapted version of Little's Personal Projects Cross-Impact Matrix (1983b). All adolescents were requested to rate to what extent their health/body goal, that was fixed as 'doing my treatment', had a positive/negative influence on their most important school/work, relationships, free time, and yourself goal. All items were rated on an 11-point Likert scale (0 = *no positive/negative influence*, 10 = *strong positive/negative influence*), with higher scores indicating a more positive/negative influence. A mean score of these four items was calculated. The adapted matrix was successfully tested on comprehensibility and relevance in a pilot study with eight healthy adolescents.

**Data analysis**

The current data had a multilevel structure as the daily reports (level 1) were hierarchically nested within the participating individuals (level 2). The level 1 variables consisted of the daily observations of positive and negative mood, goal facilitation and goal interference (21 days), whereas the level 2 variables consisted of the between-person variables gender (dummy coded: 0 = male, 1 = female), age, time since diagnosis, type of diagnosis (dummy coded: 0 = cystic fibrosis, 1 = diabetes), negative life events, goal-related self-efficacy, and acceptance (38 persons). Because of the hierarchical structure, a series of multilevel regression analyses using Hierarchical Linear Modeling (HLM 6.0 software package) was performed.

Before testing the hypotheses, baseline models without any predictors were run for each dependent variable to calculate the amount of within (level 1) and between person (level 2) variance. To test the hypotheses including predictions over time (i.e., interference/facilitation and next day mood), variables with lags of one day ($n$-1) were created for daily goal interference and facilitation. To explore whether the relationship between acceptance and daily mood is explained by goal interference and facilitation the procedure for investigating mediation described in Nezlek (2011) was followed.

All level 1 variables were entered group mean centered. Level 2 variables were entered grand mean centered, except for the categorical variables, which were entered uncentered. A random error term was estimated for each level 1 variable. Full maximum likelihood estimation was used for all analyses. Associated effect-size correlations (see Rosenthal & Rosnow, 1991, p. 441) were estimated for each set of multilevel regression analyses, with $r = .10$ indicating a small effect size, $r = .30$ indicating a medium effect size, and $r = .50$ indicating a large effect size (Cohen, 1992).

**RESULTS**

**Descriptives**

Diary data were available for 38 adolescents. A total of 737 days were completed out of a possible 798 days (completion rate = 92.36 %). *T*-tests revealed that, in comparison with adolescents with diabetes, adolescents with CF had a significant longer time since diagnosis ($M_c = 16.28$, $SD_c = 5.83$; $M_d = 8.07$, $SD_d = 4.46$; $t = 4.71$, $p = .00$). No significant differences in health status between adolescents with cystic fibrosis and diabetes were found ($M_c = 2.95$, $SD_c = 2.55$; $M_d = 3.56$, $SD_d = .81$; $t = -.92$, $p = .37$). Mean score for acceptance was 19.32 ($SD = 3.04$), which is in line with previous research in

adolescents with CF (Casier et al., 2011). Adolescents reported on average 1.11 negative life events during the past year ($SD$ = 1.09). Mean goal-related self-efficacy was 3.08 ($SD$ = .34). Mean positive mood was 16.65 ($SD$ = 4.81), whereas mean negative mood was 3.72 ($SD$ = 4.72). The mean for goal facilitation was 9.53 ($SD$ = 7.05) and 2.71 ($SD$ = 4.01) for goal interference.

**The role of acceptance in daily mood**

Forty-two per cent of the variance in negative mood was due to variation between adolescents (level 2) and 58% to variation within adolescents (level 1). Fifty-two per cent of the variance in positive mood was due to variation between adolescents (level 2) and 48% to variation within adolescents (level 1). In line with expectations, acceptance was negatively related to negative mood ($\beta_{06}$ = -.34, $t$(30) = -2.49, $p$ = .02, $r$ = .32) and positively related to positive mood ($\beta_{06}$ = .55, $t$(30) = 4.73, $p$ = .00, $r$ = .53). Of the control variables, time since diagnosis and type of diagnosis were significantly related to negative mood, and type of diagnosis also to positive mood (see Table 1). The longer the time since diagnosis, the more negative mood was reported. Furthermore, adolescents with diabetes reported on average higher levels of negative mood and lower levels of positive mood than adolescents with cystic fibrosis.

Table 1

*Multilevel regression model for daily negative and positive mood regressed on acceptance*

| | Negative mood | | | Positive mood | | |
|---|---|---|---|---|---|---|
| | Coefficient | SE | $t$ | Coefficient | SE | $t$ |
| Intercept ($\beta_{00}$) | 2.04** | .55** | 3.68** | 17.90** | .80** | 22.29** |
| Gender ($\beta_{01}$) | -.08 | .93 | -.08 | .52 | 1.00 | .52 |
| Age ($\beta_{02}$) | .02 | .02 | .79 | -.04 | .02 | -1.93 |
| Time since diagnosis ($\beta_{03}$) | .02* | .01* | 2.71* | -.01 | .01 | -1.30 |
| Type of diagnosis ($\beta_{04}$) | 3.90** | .93** | 4.22** | -3.60** | .81** | -4.39** |
| Health status ($\beta_{05}$) | -.71 | .45 | -1.60 | 1.29 | .64 | 2.01 |
| Acceptance ($\beta_{06}$) | -.34* | .14* | -2.49* | .55** | .12** | 4.73** |

Note: $Y_{ti} = \beta_{00} + \beta_{01}$(gender) + $\beta_{02}$(age) + $\beta_{03}$(time since diagnosis) + $\beta_{04}$(type of diagnosis) + $\beta_{05}$(health status) + $\beta_{06}$(acceptance) + $R_{0i} + E_{ti}$

\* $p$ < .05, \*\* $p$ < .01

**The role of acceptance in daily goal interference and facilitation**

Fifty-nine per cent of the variance in goal interference was due to variation between adolescents (level 2) and 41% to variation within adolescents (level 1). Eighty-five per cent of the variance in goal facilitation was due to variation between adolescents

(level 2) and 15% to variation within adolescents (level 1). Multilevel analyses (see Table 2) revealed that acceptance of illness was not related to experienced interference of life goals by treatment goals ($\beta_{08}$ = -.22, $t(27)$ = -1.36, $p$ = .18, $r$ = .20), nor to facilitation of life goals by treatment goals ($\beta_{08}$ = -.51, $t(24)$ = -1.28, $p$ = .21, $r$ = .20). None of the control variables (gender, age, time since diagnosis, type of diagnosis, health status, negative life events, and goal-related self-efficacy) were related to goal interference and facilitation.

Table 2

*Multilevel regression model for daily experienced goal interference and goal facilitation regressed on acceptance*

|  | Goal interference | | | Goal facilitation | | |
|---|---|---|---|---|---|---|
|  | Coefficient | SE | $t$ | Coefficient | SE | $t$ |
| Intercept ($\beta_{00}$) | 2.89** | .93** | 3.10** | 8.19** | 1.96** | 4.18** |
| Gender ($\beta_{01}$) | -1.15 | 1.31 | -.88 | 3.42 | 2.29 | 1.49 |
| Age ($\beta_{02}$) | -.04 | .03 | -1.44 | -.04 | .05 | -.78 |
| Time since diagnosis ($\beta_{03}$) | .01 | .01 | 1.20 | -.01 | .02 | -.72 |
| Type of diagnosis ($\beta_{04}$) | 1.51 | 1.13 | 1.34 | -.94 | 2.58 | -.36 |
| Health status ($\beta_{05}$) | -1.20 | .78 | -1.54 | .77 | 1.26 | .61 |
| Negative life events ($\beta_{06}$) | .52 | .53 | .98 | -.93 | .84 | -1.11 |
| Goal-related self-efficacy ($\beta_{07}$) | .99 | 2.05 | .49 | -.90 | 2.83 | -.32 |
| Acceptance ($\beta_{08}$) | -.22 | .16 | -1.36 | -.51 | .39 | -1.28 |

Note: $Y_{ti} = \beta_{00} + \beta_{01}(\text{gender}) + \beta_{02}(\text{age}) + \beta_{03}(\text{time since diagnosis}) + \beta_{04}(\text{type of diagnosis})_+ \beta_{05}(\text{health status}) + \beta_{06}(\text{negative life events}) + \beta_{07}(\text{goal-related self-efficacy}) + \beta_{08}(\text{acceptance}) + r_{0i} + e_{ti}$

\* $p < .05$, \*\* $p < .01$

**The role of goal interference and facilitation in daily mood**

Contrary to expectations (see Tables 3 and 4), higher experienced interference of life goals by treatment goals was unrelated to same day negative mood ($\beta_{10}$ = .12, $t(35)$ = 1.52, $p$ = .14, $r$ = .19), same day positive mood ($\beta_{10}$ = -.05, $t(35)$ = -.60, $p$ = .55, $r$ = .06), next day negative mood ($\beta_{10}$ = .02, $t(35)$ = .27, $p$ = .79, $r$ = .03) and next day positive mood ($\beta_{10}$ = -.04, $t(35)$ = -.48, $p$ = .63, $r$ = .06). Facilitation of life goals by treatment goals was unrelated to same day negative mood ($\beta_{10}$ = -.10, $t(32)$ = -.96, $p$ = .35, $r$ = .12), same day positive mood ($\beta_{10}$ = .16, $t(32)$ = 1.92, $p$ = .06, $r$ = .22), next day negative mood ($\beta_{10}$ = -.04, $t(32)$ = -.45, $p$ = .66, $r$ = .06) and next day positive mood ($\beta_{10}$ = .08, $t(32)$ = 1.00, $p$ = .32, $r$ = .12). Of the control variables type of diagnosis and time since diagnosis were significantly related to negative mood (see Table 3 and 4). Having diabetes and a

longer time since diagnosis were associated with higher levels of negative mood. Furthermore, age, type of diagnosis, and health status were significantly related to positive mood (see Table 3 and 4). This indicated that a higher age, having diabetes, and a worse health status were related to lower levels of positive mood.

Table 3

*Multilevel regression model for daily negative and daily positive mood regressed on goal interference and facilitation*

|  | Negative mood | | | Positive mood | | |
|---|---|---|---|---|---|---|
|  | Coefficient | SE | $t$ | Coefficient | SE | $t$ |
| Intercept ($\beta_{00}$) | 1.26* | .60* | 2.11* | 18.79** | .96** | 19.59** |
| Gender ($\beta_{01}$) | 1.35 | .96 | 1.41 | -1.30 | 1.02 | -1.28 |
| Age ($\beta_{02}$) | .04 | .02 | 1.90 | -.07** | .02** | -3.09** |
| Time since diagnosis ($\beta_{03}$) | .01* | .01 | 2.07 | -.00 | .01 | -.40 |
| Type of diagnosis ($\beta_{04}$) | 3.87** | 1.00** | 3.87** | -3.49** | 1.01** | -3.45** |
| Health status ($\beta_{05}$) | -.57 | .49 | -1.17 | 1.33 | .82 | 1.62 |
| Goal interference ($\beta_{10}$) | .12 | .08 | 1.52 | -.05 | .08 | -.60 |
| Intercept ($\beta_{00}$) | 1.02 | .67 | 1.51 | 19.07** | .94** | 20.32** |
| Gender ($\beta_{01}$) | 1.10 | .88 | 1.25 | -1.50 | .96 | -1.56 |
| Age ($\beta_{02}$) | .03 | .02 | 1.72 | -.08** | .02** | -3.99** |
| Time since diagnosis ($\beta_{03}$) | .02* | .01* | 2.72* | -.00 | .01 | .10 |
| Type of diagnosis ($\beta_{04}$) | 4.32** | 1.03** | 4.18** | -4.01** | .80** | -4.98** |
| Health status ($\beta_{05}$) | -.45 | .47 | -.97 | 1.36 | .79 | 1.72 |
| Goal facilitation ($\beta_{10}$) | -.10 | .11 | -.96 | .16 | .08 | 1.92 |

Note: $Y_{ti} = \beta_{00} + \beta_{01}(\text{gender}_i) + \beta_{02}(\text{age}_i) + \beta_{03}(\text{time since diagnosis}_i) + \beta_{04}(\text{type of diagnosis}_i) + \beta_{05}(\text{health status}_i) + \beta_{10}(\text{goal interference}_{ti}) + r_{0i} + r_{1i}(\text{goal interference}_{ti}) + e_{ti}$ or $Y_{ti} = \beta_{00} + \beta_{01}(\text{gender}_i) + \beta_{02}(\text{age}_i) + \beta_{03}(\text{time since diagnosis}_i) + \beta_{04}(\text{type of diagnosis}_i) + \beta_{05}(\text{health status}_i) + \beta_{10}(\text{goal facilitation}_{ti}) + r_{0i} + r_{1i}(\text{goal facilitation}_{ti}) + e_{ti}$

* $p < .05$, ** $p < .01$

Table 4

*Time-lagged multilevel regression model for next day negative and positive mood regressed on daily goal interference and facilitation*

| | Next day negative mood | | | Next day positive mood | | |
|---|---|---|---|---|---|---|
| | Coefficient | SE | $t$ | Coefficient | SE | $t$ |
| Intercept ($\beta_{00}$) | 1.66* | .65* | 2.57* | 18.78** | .99** | 19.06** |
| Gender ($\beta_{01}$) | .66 | 1.06 | .63 | -1.23 | 1.07 | -1.15 |
| Age ($\beta_{02}$) | .03 | .02 | 1.11 | -.06* | .02* | -2.71* |
| Time since diagnosis ($\beta_{03}$) | .02* | .01* | 2.22* | -.00 | .01 | -.48 |
| Type of diagnosis ($\beta_{04}$) | 3.96** | 1.11** | 3.57** | -3.90** | 1.03** | -3.78** |
| Health status ($\beta_{05}$) | -.85 | .53 | -1.59 | 1.54 | .85 | 1.80 |
| Goal interference ($\beta_{10}$) | .02 | .09 | .27 | -.03 | .08 | -.48 |
| Intercept ($\beta_{00}$) | 1.31 | .76 | 1.71 | 19.07** | .94** | 20.30** |
| Gender ($\beta_{01}$) | .31 | 1.29 | .24 | -.66 | 1.01 | -.65 |
| Age ($\beta_{02}$) | .01 | .03 | .40 | -.06* | .02* | -2.70* |
| Time since diagnosis ($\beta_{03}$) | .03* | .01* | 2.80* | -.01 | .01 | -2.03 |
| Type of diagnosis ($\beta_{04}$) | 4.94** | 1.26** | 3.92** | - 5.15** | .77** | -6.68** |
| Health status ($\beta_{05}$) | -.93 | .55 | -1.69 | 1.77* | .78* | 2.26* |
| Goal facilitation ($\beta_{10}$) | -.04 | .08 | -.45 | .08 | .08 | 1.00 |

Note: $Y_{ti} = \beta_{00} + \beta_{01}(\text{gender}_i) + \beta_{02}(\text{age}_i) + \beta_{03}(\text{time since diagnosis}_i) + \beta_{04}(\text{type of diagnosis}_i) + \beta_{05}(\text{health status}_i) + \beta_{10}(\text{goal interference}_{ti}) + r_{0i} + r_{1i}(\text{goal interference}_{ti}) + e_{ti}$ or $Y_{ti} = \beta_{00} + \beta_{01}(\text{gender}_i) + \beta_{02}(\text{age}_i) + \beta_{03}(\text{time since diagnosis}_i) + \beta_{04}(\text{type of diagnosis}_i) + \beta_{05}(\text{health status}_i) + \beta_{10}(\text{goal facilitation}_{ti}) + r_{0i} + r_{1i}(\text{goal facilitation}_{ti}) + e_{ti}$

\* $p < .05$, \*\* $p < .01$

## DISCUSSION

To our knowledge, this is the first study using a daily diary approach that investigates the relationships between acceptance, daily well-being, goal interference and facilitation by treatment goals in adolescents with chronic illness. It was found that higher acceptance related to better daily well-being, but not to interference of life goals by treatment goals, nor to facilitation. Furthermore, goal interference and facilitation were unrelated to daily well-being.

In line with previous research (e.g., Casier et al., 2011), higher levels of acceptance were found to be related to better daily well-being. Innovative in this study was the daily process approach. Our finding that acceptance explained daily mood in adolescents with chronic illness is in line with the conceptualisation of acceptance. Acceptance has been described as the strength to focus on what is pleasurable in life despite being ill. Instead of a sign of weakness, resignation, and failure, it is seen as a method to increase action toward what is valued in life (Evers et al., 2001; Hayes et al.,

2006; Risdon et al., 2003). By shifting attention from illness to valuable domains of life, a meaningful life and well-being can be sustained (Hayes et al., 2006).

This study also, for the first time, investigated the relationship between acceptance and goal processes such as the experience of interference (when striving for one goal decreases the chance of attaining another goal) or facilitation (when striving for one goal increases the chance of attaining another goal) between goals. Contrary to our expectations, acceptance was not related to any of these experiences. Possible explanation for this lack of findings is that acceptance of illness may not relate to the experience of goal interference and facilitation per se, but rather to the way one deals with these experiences. This explanation is in line with the conceptualisation of acceptance, defined in terms of 'adapting to chronic illness' and 'handling averse consequences' (Evers et al., 2001, p. 1027). It suggests that acceptance may rather be associated with other goal-related constructs such as a balance between treatment goals and other important life goals, the relative importance of these goals, and the amount of success in the pursuit of these goals. Acceptance indeed includes an engagement in important life goals despite being ill (Evers et al., 2001; Hayes et al., 2006). This may mean that a balanced interplay between treatment goals (e.g., doing treatment) and other important life goals (e.g., building social relationships, getting an academic degree, becoming independent) is essential. Future research should investigate if acceptance indeed relates to these constructs and experiences.

Furthermore, the current findings revealed that the previously established relationship between goal interference and well-being (for an overview see Riediger, 2007) has not been confirmed in the context of chronic illness and/or when studied from a daily process perspective. Several explanations can be proposed for these unexpected results. First of all, goal interference may not have instant but rather long-term effects on well-being (Boersma, Maes, & van Elderen, 2005). The present study does not allow conclusions about possible long-term effects. Future research should therefore investigate this association within a broader time frame (e.g., weeks or months instead of days). Second, it is possible that goal interference may only relate to daily well-being in a subsample. Given that (a) adolescents with diabetes reported worse mood but equal levels of goal interference compared to adolescents with CF and (b) adolescents with CF, compared to adolescents with diabetes, already had more time to find a way to deal with the experience of goal interference (i.e., longer time since diagnosis), it is possible that the type of diagnosis moderates the relationship between goal interference and daily well-

being. Further research using larger samples is needed to investigate this hypothesis. Ways of handling the experience of goal interference may include a search for compatible strategies to attain different goals, the identification of possible instrumental relations between goals, the optimisation of the use of limited resources (e.g., time, energy), the search for other goals in the same domain or alternative routes to goals, scaling back goals, or even the disengagement from incompatible and reengagement in other compatible goals (e.g., choosing home-based instead of outdoor leisure activities that can be performed during aerosol treatment). Third, not the experience of goal interference itself, but rather the emotional reactions arising from this interference may affect well-being. This explanation is in line with recent research suggesting that not the actual number of blocked goals but the frustration arising from it relates to well-being (Massey et al., 2009a; Massey, Garnefski, Gebhardt, & van der Leeden, 2009b). Fourth, the current study fixed the treatment goal of the participating adolescents as 'doing treatment'. This goal may have been formulated too general and comprehensive. Further research focussing on specific aspects of treatment that adolescents identify as very important is needed.

Next to focussing on goal interference, this study also, as a first, explored the relationship between goal facilitation and well-being in the context of chronic illness. Previous research on goal facilitation revealed mixed results, with facilitation predominantly being unrelated, but sometimes positively related to well-being (Riediger, 2007; Riediger & Freund, 2004). The current results support the findings that goal facilitation by treatment goals is unrelated to daily mood. This indicates that the degree to which the pursuit of treatment goals makes the pursuit of other valued goals easier does not relate to the degree of negative and positive mood in adolescents with CF and diabetes. That goal facilitation did not relate to mood may be explained by the fact that individuals generally do not react strongly to gains (see Riediger, 2007). For future research, comparing experienced goal interference and facilitation between individuals with chronic illness and healthy peers may be interesting. This kind of research may clarify whether it is essential to further address goal interference in chronically ill adolescents, and may offer important starting points for intervention.

Some limitations of this study should be mentioned. First, the current sample reported on average rather high levels of acceptance and low levels of goal interference. Therefore, it is uncertain whether the current findings are generalizable to adolescents reporting lower levels of acceptance and/or higher goal interference. Second, the current

study only relied on self-report measures. A multi-method approach including reports of other informants such as parents and caregivers, would contribute to the strength of the current findings. Third, acceptance was assessed with only one scale. There is a need for more instruments assessing acceptance in the context of chronic illness. These instruments should not only focus on dealing with uncontrollability, unpredictability, and negative consequences of being ill, but should also address other aspects of acceptance such as dealing with unwanted thoughts and feelings and engaging in activities that serve important life goals despite being ill (Casier et al., 2011). A starting point for the development of these instruments could be the Acceptance and Action Questionnaire (AAQ; Hayes et al., 2004). Fourth, the current results should be interpreted with caution as the response rate was rather low. It is possible that participating adolescents had particular characteristics that do not generalize to non-participating adolescents (e.g., better health status).

The findings from the current study suggest important target points for intervention in adolescents with chronic illness. As acceptance relates to daily well-being, adolescents with chronic illness may benefit from psychological interventions to promote acceptance. Approaches addressing acceptance, such as Acceptance and Commitment Therapy (ACT; Hayes, 2004) already exist for several paediatric chronic conditions (e.g., sickle cell disease, chronic pain) and demonstrate promising results (e.g., Masuda et al., 2011; Wicksell et al., 2009). Further research into the factors that may relate to and influence acceptance is needed to determine if these interventions are useful for adolescents with CF and diabetes. If acceptance indeed seems to be a workable key factor in the well-being of these adolescents, existing interventions such as ACT (Hayes, 2004) could be modified for and tested in the context of CF and diabetes.

**REFERENCES**

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14,* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Boersma, S.N., Maes, S., & van Elderen, T. (2005). Goal disturbance predicts health-related quality of life and depression 4 months after myocardial infarction. *British Journal of Health Psychology, 10,* 615-630. doi: 10.1348/135910705X52525

Bolger, N., Davis, A., & Rafaeli, E. (2003). Diary methods: capturing life as it is. *Annual Review of Psychology, 54,* 579-616. doi: 10.1146/annurev.psy.54.101601.145030

Bourke, S. J., Doe, S. J., Gascoigne, A. D., Heslop, K., Fields, M., Reynolds, D., & Mannix, K. (2009). An integrated model of provision of palliative care to patients with cystic fibrosis. Palliative Medicine, 23, 512–517. doi: 10.1177/0269216309106312

Caes, L., Vervoort, T., Eccleston C., & Goubert, L. (medium revision). Parents who catastrophize about their child's pain prioritize attempts to control pain. *Pain.*

Casier, A., Goubert, L., Huse, D., Theunis, M., Franckx, H., Robberecht, E. Matthys, D., & Crombez, G. (2011). Acceptance and well-being in adolescents and young adults with cystic fibrosis: a prospective study. *Journal of Pediatric Psychology, 36,* 476-487. doi: 10.1093/jpepsy/jsq111

Cohen, J. (1992). A power primer. Psychological Bulletin, 112, 155-159.

De Ridder, D., & Kuijer, R. (2007). Reconsidering illness-related goals: is discrepancy resolved by confronting emotions? *Psychology and Health, 22,* 107-122. doi: 10.1080/14768320600603471

Diener, E., & Emmons, R.A. (1985). The independence of positive and negative affect. *Journal of Personality and Social Psychology, 47,* 1105-1117.

Evers, A.W.M., Kraaimaat, F.W., van Langenveld, W., Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Feinstein, A.B., Forman, E.M., Masuda, A., Cohen, L.L., Herbert, J.D., Moorthy, L.N., & Goldsmith, D.P. (2011). Pain intensity, psychological inflexibility, and acceptance of pain as predictor of functioning in adolescents with juvenile idiopathic arthritis: a preliminary investigation. *Journal of Clinical Psychology in Medical Settings, 18,* 291-298. doi: 10.1007/s10880-011-9243-6

Glasscoe, C. A., & Quittner, A. L. Psychological interventions for people with cystic fibrosis and their families. Cochrane Database of Systematic Reviews 2008, Issue 3. Art. No.: CD003148. doi: 10.1002/14651858.CD003148.pub2.

Gebhardt, W.A. (2008). On multiple goals and continuous conflict. *The European Health Psychologist, 10,* 25-28.

Hayes, S.C. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapies. *Behavior Therapy, 35,* 639-665. doi: 10.1016/S00005-7894(04)80013-3

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: model, processes and outcomes. *Behaviour Research and Therapy, 44,* 1-25. doi:10.1016/j.brat.2005.06.006

Huijnen, I.P.J., Verbunt, J.A., Roelofs, J., Goossens, M., & Peters, M. (2009). The disabling role of fluctuations in physical activity in patients with chronic low back pain. *European Journal of Pain, 13,* 1076-1079. doi: 10.1016/j.ejpain.2008.12.008

Kraaij, V., Garnefski, N., de Wilde, E.J., Dijkstra, A., Gebhardt, W., Maes, S., & ter Doest Laura (2003). Negative life events and depressive symptoms in late adolescence: bonding and cognitive coping as vulnerability factors? *Journal of Youth and Adolescence, 32,* 185-193.

Lauwerier, E., Crombez, G., Van Damme, S., Goubert, L., Vogelaers, D., & Evers, A. W. M. (2010). The construct validity of the Illness Cognition Questionnaire: the robustness of the three-factor structure across patients with chronic pain and chronic fatigue. *International Journal of Behavioral Medicine, 17,* 90-96. doi: 10.1007/s12529-009-9059-z

Little, B. R. (1983a). Personal projects: A rationale and method for investigation. *Environment and Behavior, 1*5, 273–309.

Little, B.R. (1983b) Personal projects Analysis. Retrieved from http://www.brianrlittle.com/Topics/research/assessment-tools/

Maes, S. & Karoly, P. (2005). Self-regulation assessment and intervention in physical health and illness: a review. *Applied Psychology: An International Review, 54,* 267-299.

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,* 421-460. doi: 10.1016/j.dr.2008.03.002

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2009a). Self-generated goals and goal process appraisals: Relationships with sociodemographic factors and well-being. *Journal of adolescence, 32,* 501-518. doi: 10.1016/j.adolescence.2008.07.003

Massey, E.K., Garnefski, N., Gebhardt, W.A., & van der Leeden, R. (2009b). Daily frustration, cognitive coping and coping efficacy in adolescent headache: a daily diary study. *Headache, 49,* 1198-1205. doi: 10.1111/j.1526-4610.2009.01492.x

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: acceptance and commitment therapy for pediatric sickle cell disease. *Journal of pediatric Psychology, 36,* 398-408. doi: 10.1093/jpepsy/jsq118

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents with chronic pain: validation of an adapted assessment instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi: 10.1016/j.ejpain.2009.05.002

Nezlek, J.B. (2011). *Multilevel Modelling for Social and Personality Psychology.* London, UK: Sage Publications Ltd.

Pinquart, M., & Shen, Y. (2011). Behavior problems in children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 36,* 1003-1016. doi: 10.1093/jpepsy/jsr042

Pinquart, M., & Teubert, D. (2012). Academic, physical, and social functioning of children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 37,* 376-389. doi: 10.1093/jpepsy/jsn06

Riediger, M. (2007). Interference and facilitation among personal goals: Age-group differences and associations with well-being and behavior. In B.R. Little, K. Salmela-Aro, J.-E. Nurmi, & S.D. Philipps (Eds.), *Personal project pursuit: Goals action, and human flourishing* (pp. 119-143). Erlbaum.

Riediger, M. & Freund, A.M. (2004). Interference and facilitation among personal goals: differential associations with subjective well-being and persistent goal pursuit. *Personality and Social Psychology Bulletin, 30,* 1511-1523. doi: 10.1177/0146167204271184

Risdon, A., Eccleston, C., Crombez, G., & McCracken, L.M. (2003). How can we learn to live with pain? A *Q*-methodological analysis of the diverse understandings of acceptance of pain. *Social Science and Medicine, 56,* 375-386. doi: 10.1016/S0277-9536(02)00043-6

Rosenthal, R. & Rosnow, R.L. (1991). Essentials of behavioural research: Methods and data analysis (2nd e.d.). New York: McGraw-Hill.

Schwartz, L.A. & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 393-405. doi: 10.1007/s10880-006-9041-8

Schwartz, L.A. & Drotar, D. (2009). Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 34,* 954-965. doi: 10.1093/jpepsy/jsn144

Seiffge-Krenke, I. (2001). *Diabetic adolescents and their families: stress, coping, and adaptation.* Cambridge, UK: Cambridge University Press.

Vervoort, T., Huguet, A., Verhoeven, K., & Goubert, L. (2011). Mothers' and fathers' responses to their child's pain catastrophizing and disability. *Pain, 152,* 786-793. doi: 10.1016/j.pain.2010.12.010

Wicksell, R.K., Melin, L., Lekander, M., & Olsson, G.L. (2009). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – a randomized controlled trial. *Pain, 141,* 248-257. doi: 10.1016/j.pain.2008.11.006

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi: 10.1016/j.pain.2011.09.003.

# CHAPTER 6

## ACCEPTANCE, WELL-BEING, AND GOALS IN ADOLESCENTS WITH CHRONIC ILLNESS: A LONGITUDINAL STUDY[1]

**ABSTRACT**

*Objective:* This longitudinal study examined whether acceptance was associated with the well-being of adolescents and young adults with chronic illness and whether this association is mediated by interference and facilitation of life goals by treatment goals.

*Method:* Thirty participants with cystic fibrosis or diabetes filled out a battery of questionnaires measuring acceptance, negative life events, goal-related self-efficacy, well-being, and interference and facilitation of life goals by treatment goals at three points in time (baseline, two and four months later).

*Results:* It was found that acceptance at time 1 was positively associated with some dimensions of well-being at time 3 (i.e., anxiety and depressive symptoms, but not health-related quality of life), but not associated with daily goal interference and facilitation at time 2. Also, there was no association between daily goal interference and facilitation at time 2 and all aspects of well-being at time 3.

*Conclusion:* The current results indicate that acceptance of illness may impact upon experienced anxiety and depressive symptoms of adolescents with cystic fibrosis or diabetes. This association could not be explained by daily experienced interference and facilitation of life goals by treatment goals. Further research is needed to identify the mechanisms underlying the association between acceptance and well-being.

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## INTRODUCTION

It is commonly known that chronic illness can seriously threaten the well-being of adolescents and young adults. Well-being includes a broad area of experiences that encompasses positive and negative affect, general life satisfaction, and satisfaction with specific domains of life (Diener, 1984; Diener, Suh, Lucas, & Smith, 1999). Indeed, in previous research the prolonged course and non-spontaneous resolution of illness, together with the improbability of a complete cure and functional impairment or disability, has repeatedly been associated with impairments in adolescents' and young adults' academic, emotional, physical, as well as social functioning (Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Yeo & Sawyer, 2005).

Despite the potentially adverse effects of growing up with a chronic illness, many adolescents and young adults with chronic illness are functioning well (Pinquart & Teubert, 2012; Yeo & Sawyer, 2010). As well-being considerably varies across affected individuals (regardless illness severity and duration), identifying factors that may explain this variation is essential. A factor that may be determinative in their well-being is 'acceptance of illness' (Evers et al., 2001; Maes & Karoly, 2005). Acceptance is conceptualized as 'recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences' (Evers et al., 2001, p. 1027). It refers to the ability to harmonise with the limitations the disease involves and to acknowledge the constantly changing demands of the disease, while staying engaged in what one finds important in life (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003). Indeed, research is accumulating that acceptance may have positive effects on the well-being of adolescents and young adults with chronic illnesses such as chronic pain (e.g., McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011), juvenile arthritis (Feinstein et al., 2011), sickle cell disease (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011), and cystic fibrosis (e.g., Casier et al., 2008, 2011). It was found that higher levels of acceptance are related to several indicators of well-being such as less anxiety and depression, better mood, better emotional, social, and physical functioning, and less disability.

Despite extensive research into the relationship between acceptance and well-being, no research has yet investigated processes underlying this relationship. The current study focusses on the experience of interference and/or facilitation of life goals (e.g., becoming independent) by treatment goals as possible mediating processes. Previous

research has shown that chronic illness may seriously affect the attainment of important life goals. This because it imposes the pursuit of new goals related to treatment in response to this illness (Badlan, 2006; Schwartz & Drotar, 2006; Seiffge-Krenke, 1998). In this context, goal interference emerges when striving for treatment goals reduces the possibility of achieving other goals and can be attributed to limited resources (e.g., time, energy) and/or incompatible goal strategies (e.g., 'adhere to treatment' and 'not let illness predominate life') (Little, 1983; Riediger, 2001; Riediger & Freund, 2004). Goal facilitation then emerges when the pursuit of treatment goals enhances the possibility of achieving another goal and is due to instrumental relations between goals and/or compatible goal strategies (e.g., 'find a routine in doing treatment' and 'make enough time for leisure activities) (Little, 1983; Riediger, 2001; Riediger & Freund, 2004). As acceptance is conceptualised as a way to engage in important life goals although being ill, it is indeed possible that acceptance is linked to how treatment and other life goals interfere with and/or facilitate each other (Evers et al., 2001; Hayes, Luoma, Bond, Masuda, Lillis, 2006; Maes & Karoly, 2005; Risdon, Eccleston, Crombez, & McCracken, 2003). Furthermore, as previous research has shown that goal interference and facilitation relates to well-being in healthy samples, this may also be the case in chronic illness (Riediger, 2007; Riediger & Freund, 2004). Whether goal interference and facilitation relate to well-being in adolescents with chronic illness, has yet to be investigated.

Given that chronic illness may impact upon goal regulation and related well-being of affected individuals, it is necessary to investigate how successful regulation of treatment and other personal goals and well-being can be achieved despite being ill. This is especially important during adolescence and young adulthood as they need to achieve important life goals to pass on to adulthood, start to regulate their goals more autonomously, are cognitively capable to evaluate the impact of illness on goal regulation, have a strong desire to be 'normal', and tend to be non-adherent to treatment (Massey, Gebhardt, & Garnefski, 2008; Nurmi, 1987, 1991,1993; Schwartz & Drotar, 2009). Aims of the current study are therefore to longitudinally investigate whether the relationship between acceptance of illness measured at time 1 (baseline) and well-being at time 3 (4 months) is mediated by goal interference/facilitation by treatment goals at time 2. This study focused on adolescents with cystic fibrosis (CF) and diabetes because both illnesses require a strict daily treatment regimen (Badlan, 2006; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009; Seiffge-Krenke, 2001). Well-being was operationalized in terms of anxiety and depressive symptoms, and health-related quality of life, as these

are core components of well-being, and can be detrimentally affected in adolescents with chronic illness (Diener et al., 1999; Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Yeo & Sawyer, 2005). In line with expert consensus, health-related quality of life was operationalized as a subjective construct that fundamentally includes four core domains: physical functioning, emotional functioning, social functioning, and role functioning (Cella, 1998; Spilker, 1996). Furthermore, we controlled for negative life events and goal-related self-efficacy as these variables may affect goal pursuit (Schwartz & Drotar, 2009). We hypothesized that (a) more acceptance is related to higher well-being, (b) more acceptance is related to less goal-interference and more goal facilitation of life goals by treatment goals, that (c) less goal-interference and more goal facilitation of life goals is related to higher well-being, and (d) that the relationship between acceptance and well-being is mediated by goal interference and facilitation.

## METHOD

The current study is part of a larger one, i.e. the Goals, Acceptance, and Well-being in Chronic Illness (GAWCI) study. The GAWCI study was conducted between September 2010 and April 2012 in Flanders, Belgium. Adolescents and young adults with CF and diabetes and matched (age and gender) healthy peers were selected for this study. The healthy peers were recruited from a sample of school children who participated between 2008 and 2011 in one of two previously conducted studies and had given consent to be re-contacted for other studies (Caes, Vervoort, Eccleston, & Goubert, 2012; Vervoort, Huguet, Verhoeven, & Goubert, 2011). These healthy adolescents participated in another part of the GAWCI study reported elsewhere. All of four Flemish University Hospitals were contacted to recruit the adolescents and young adults with CF. Two of them, the University Hospitals of Ghent and Leuven, agreed to participate. For the adolescents and young adults with diabetes, the University Hospital of Ghent was contacted, who agreed to participate.

### Participants

All participants needed to meet the following criteria: 14-22 years of age, understanding the Dutch language, and no developmental disorder. For the adolescents and young adults with CF an additional inclusion criterion stating that they should not have planned/underwent a lung transplantation was used. This was done because of the exceptional status of this situation (e.g., very high level of symptoms, being terminally ill). One hundred and thirty adolescents and young adults (CF = 74, diabetes = 56), met

these criteria. These adolescents were screened by the psychologist from the respective hospital. All adolescents eligible for participation received an invitation letter and/or were recruited during one of their hospital visits by their psychologist (time period of recruitment: September 2010-April 2011). Adolescents who were interested to participate ($N$ = 82) where contacted by phone by a research assistant to arrange a home visit. Eventually, 61 of the 130 eligible adolescents and young adults (CF: $N$ = 35, 17 males, 18 females; diabetes: $N$ = 26, 10 males, 16 females) were enrolled in our study (response rate = 46.92%). The main reasons for not participating were a lack of time and interest. Of these adolescents and young adults, 30 took part at all three time points (participation rate = 49.18%; CF: $N$ = 14, 10 males, 4 females; diabetes: $N$ = 16, 7 males, 9 females). All adolescents were of Caucasian origin. Mean age was 19.17 years ($SD$ = 2.87) for the adolescents and young adults with CF and 15.65 years ($SD$ = 1.16) for the adolescents and young adults with diabetes. Mean time since diagnosis was 17.04 years ($SD$ = 5.58) for the adolescents and young adults with CF, and 7.54 years ($SD$ = 4.37) for the adolescents and young adults with diabetes. Adolescents and young adults with CF were significantly older ($F(1,29)$ = 20.43, $p$ = .00) and had a significant longer time since diagnosis ($F(1,28)$ = 26.24, $p$ = .00) compared with adolescents and young adults with diabetes. Regarding health status no significant differences between participants with CF and diabetes were found ($M_c$ = 3.64, $SD_c$ = .63; $M_d$ = 3.80, $SD_d$ = .86; $F(1,28)$ = .31, $ns$). Of the eligible adolescents who did not participate, 60% was diagnosed with CF and 40% with diabetes. No further information about the non-participants was available due to confidentiality.

**Procedure**

At time 1, a research assistant visited the participants at their home. During this visit, written parent consent (adolescents < 18 years) and/or adolescent assent (adolescents < 18 years)/consent (adolescents > 18 years) were obtained, and a booklet of questionnaires was filled out. At 2 (time 2) and 4 months (time 3), all adolescents and young adults were contacted again and asked to complete the booklet of questionnaires a second and third time. All follow-up questionnaires were sent by regular mail. Forty of the 61 participants (65.57%) returned the Time 2 questionnaires, whereas 30 of them (49.18%) returned the questionnaires at Time 3. This study was approved by the ethical committees of the University Hospitals of Ghent and Leuven.

**Measures**

*Socio-demographic information* (age, gender, time since diagnosis, type of diagnosis) was verbally inquired before filling out the questionnaires.

*Acceptance* was assessed by the acceptance scale of the Dutch version of the Illness Cognition Questionnaire (ICQ; Evers et al., 2001). Acceptance is conceptualised as recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences (six items, e.g. "I have learned to live with my illness"). Four-point Likert scales are used to rate all items (1 = *not at all*, 4 = *completely*). Total scores range from 6 to 24, with higher scores denoting higher levels of acceptance. Psychometric qualities of the ICQ were proven adequate in samples of Dutch-speaking adults with rheumatoid arthritis, multiple sclerosis, chronic pain and chronic fatigue (Evers et al., 2001; Lauwerier et al., 2010). Reliability of the acceptance scale in the research sample of the present study was good with a Cronbach's α of .80.

*Goals* were assessed with an open goal-elicitation procedure. To cover all important domains of life, participants were requested to report their goals in the domains of school/work, relationships, leisure time, personal development, and health/body (for a review, see Massey, Gebhardt, & Garnefski, 2008). Participants could report as many goals as desired. In line with previous research (Massey, Gebhardt, & Garnefski, 2009a), goals were elicited as following: 'Some things you find important in life, other things you find less important. People differ in this respect: what one considers important and consequently has as a goal, can be less important for another. Everyone has his or her own goals, and that makes every person unique and special. Some goals you like to attain, other goals matter less. We would like to ask you some questions concerning the things you consider important in life. In other words: what are your most important goals? It is important to know that : (a) one can have different goals in different domains at the same time, for example in the domain of school, relationships, work, yourself, leisure time, or health, (b) goals have to do with either wanting to do something or exactly not wanting to do it. We are interested in the goals that you are currently pursuing, and that will still be important for you within a few months.'. After elicitation, every participant selected his/her most important goal per domain, except for the health/body domain. Because this study aimed at investigating the interaction between personal goals and treatment goals, the goal 'doing my treatment' was fixed as most important within the health/body domain. Before administration, this procedure was pilot tested on comprehensibility and

relevance in eight healthy adolescents and young adults. This pilot test was successful, so no adjustments to the procedure were made.

*Goal interference and facilitation* were measured using an adapted version of Little's Personal Projects Cross-Impact Matrix (1983). New in this version was that interference and facilitation were measured as two orthogonal dimensions instead of two ends of the same dimension. All participants rated to what extent their fixed health/body goal (i.e., 'doing my treatment'), had a positive/negative influence on their selected school/work, relationships, free time, and personal development goal. An 11-point Likert scale was used to rate these items (0 = no positive/negative influence, 10 = strong positive/negative influence) and a mean score of all items was calculated. Higher scores denoted a more positive/negative influence. The modified matrix was successfully piloted on comprehensibility and relevance in eight healthy adolescents and young adults.

*Anxiety and depressive symptoms* were assessed by the Dutch version of the Hospital Anxiety and Depression Scale (HADS; Spinhoven et al., 1997; Zigmond & Snaith, 1983). The HADS consists of 14 items to be rated on a 4-point Likert scale, and has two subscales: 'anxiety' (7 items, e.g. "Do you feel tense and wound up?") and 'depression' (7 items, e.g. "Do you feel cheerful?"). Total scores range between 0 and 21, with higher scores indicating higher levels of anxiety and/or depressive symptoms (Snaith, 2003). For depression, scores between 7 and 9 are indicative of possible depression, scores above 9 of probable depression. Cut-off scores for anxiety are 9 (possible emotional disorder) and 12 (probable emotional disorder) (White, Leach, Sims, Atkinson, & Cottrell, 1999). The HADS is designed for use in medical practice. It is proven reliable and valid as a screening instrument in adolescents, adults, and elderly subjects with or without a medical condition (Spinhoven et al., 1997; White et al., 1999; Zigmond & Snaith, 1983). Cronbach's α in this study was .74 for anxiety and .61 for depression at time 3 (see Table 1).

*Quality of life* was assessed by the Pediatric Quality of Life Inventory™ Version 4.0 (PedsQL; Varni, 1998). This 23-item inventory measures the core dimensions of health: physical functioning (8 items, e.g., "It's hard for me to run."), emotional functioning (5 items, e.g., "I feel sad or blue."), social functioning (5 items, e.g., "It is difficult to keep up with peers."), and work/school functioning (5 items, e.g., "I forget things."). The instructions inquire how much of a problem each item has been during the past 1 month. All items are rated on a 5-point Likert scale (0 = never a problem, 4 = almost always a problem). Items are reverse-scored and linearly transformed to a 0–100

scale, with higher scores indicating better health-related quality of life. The 4 scales can be combined to 3 summary scores: a total scale score (23 items), a physical health summary score (8 items) and a psychosocial health summary score (15 items). The summary scores are computed as the sum of the items divided by the number of items answered in the corresponding subscales. In this study the physical and psychosocial health summary scores were used. The PedsQL has proven reliable, valid (i.e., distinguishes healthy individuals from individuals with health conditions and distinguishes according to disease severity), and responsive to clinical change over time (Varni, 1998-2011; see http://www.pedsql.org/about_pedsql.html). Reliability (Cronbach's α) of the PedsQL in the current sample was .91 for physical health and .91 for psychosocial health (see Table 1).

*Negative life events* were assessed by a 12-item lifetime checklist for adolescents (Kraaij et al., 2003). This list focuses on negative life events regarding self or significant others during the past year, such as death of someone close, parental divorce, and abuse experiences. Total scores (range 0-12) are obtained by adding up all negative life events experienced.

*Goal-related self-efficacy* is measured as the perception of self-efficacy in relation to being able to achieve one's goals despite possible obstacles by an adaptation of Schwarzer's General Self-Efficacy Scale (Massey et al., 2008; Schwarzer & Jerusalem, 1995). This scale has 10 items (e.g., "I can always manage to achieve my goals if I try hard enough") that need to be scored on a 4-point Likert scale (1 = not at all true, 4 = exactly true). Total scores were the mean score of these 10 items (range 1-4). Higher scores denoted more goal related self-efficacy. Cronbach's α in the current sample was .82.

*Health status* was subjectively rated on a 5-point Likert scale ("How would you describe your current health status?"; 1 = bad, 5 = excellent).

**Data analysis**

All analyses were performed using SPSS 20.0. Descriptive statistics (means, *SD*'s, Cronbach's α) were computed for all variables of interest. Pearson correlations were performed to determine whether gender, age, time since diagnosis, type of diagnosis, health status, negative life events, and goal-related self-efficacy were related to the well-being and goal measures (see Table 1). To test for mediation, the indirect effect of acceptance on well-being through goal interference and facilitation was measured using the nonparametric bootstrapping method described by Preacher and Hayes (2004)

was used (also see http://afhayes.com/spss-sas-and-mplus-macros-and-code.html). This method has increasingly been used and is recommended for studies with a smaller sample size (Hayes, 2009; MacKinnon, Lockwood, Hoffman, West, & Sheets, 2002; Shrout & Bolger, 2002). To test for mediation, several effects and their weights need to be differentiated (see Figure 1). There is a total effect of acceptance on well-being (weight *c*), that consists of (1) the direct effect of acceptance on well-being (weight *c'*) and (2) the indirect effect of acceptance on well-being through a mediator, i.e. goal interference or facilitation (weight *ab*). By using this method 95% bias-corrected and accelerated bootstrap confidence intervals for the indirect effects were generated, using 5000 bootstrap samples. Separate mediation models were tested for each outcome variable (i.e., anxiety symptoms, depressive symptoms, physical health, and psychosocial health). In line with the recommendations of Cole and Maxwell (2003), data on the independent variable, mediators, and outcome variables were collected at three subsequent points in time (time 1, time 2, and time 3).
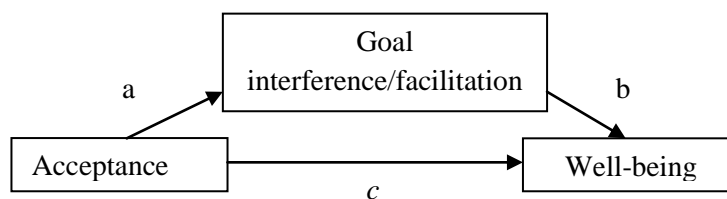


*Note.* The total effect (weight c) consists of a direct effect (weight c') and the indirect effect (ab weight).

*Figure 1.* Graphic representation of the mediation model

**RESULTS**

**Descriptives**

Mean scores, standard deviations (*SD*'s), internal consistencies (Cronbach's α) and correlations for/between all variables are presented in Table 1. The mean score for acceptance was comparable to the levels of acceptance in previous studies in adolescents with CF (Casier et al., 2008, 2011). All adolescents scored beneath the cut-off for probable depression, and 93.30% scored beneath the cut-off for probable anxiety. These scores reflect low psychological disturbance.

Table 1

*Means (M), standard deviations (SD), internal consistency (Cronbach's α) of age, gender, time since diagnosis, type of diagnosis, health status, negative life events, goal-related self-efficacy, acceptance (time 1), goal interference (time 2), goal facilitation (time 2), anxiety symptoms (time 3), depressive symptoms (time 3), physical health (time 3), and psychosocial health (time 3)*

| | M | SD | α | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1. Age | 17.29 | 2.75 | - | -.28 | .80** | -.65** | .02 | .42* | .35 | .01 | .10 | -.04 | -.05 | .16 | -.38* | -.24 |
| 2. Gender | - | - | - | - | -.29 | .28 | -.13 | -.07 | -.53** | -.30 | .04 | .34 | .37* | .08 | .06 | -.14 |
| 3. Time since diagnosis | 12.12 | 6.88 | - | - | - | -.70** | -.06 | .38* | .47* | .08 | .18 | -.25 | -.09 | .10 | -.39* | -.18 |
| 4. Type of diagnosis | - | - | - | - | - | - | .11 | -.50** | -.32 | -.28 | -.03 | .26 | .20 | .02 | .33 | .11 |
| 5. Health status | 3.72 | 2.62 | - | - | - | - | - | -.59** | .01 | .19 | -.36 | -.04 | -.29 | -.38* | .42* | .47* |
| 6. Negative life events | 1.00 | .95 | - | - | - | - | - | - | .11 | .07 | .21 | -.01 | .23 | .08 | -.37* | -.37* |
| 7. Goal-related self-efficacy | 3.03 | .34 | .82 | - | - | - | - | - | - | .39* | -.08 | -.18 | -.22 | -.13 | -.18 | -.05 |
| 8. Acceptance (time 1) | 18.70 | 2.97 | .80 | - | - | - | - | - | - | - | -.25 | -.22 | -.56** | -.55** | .11 | .41* |
| 9. Goal interference (time 2) | 2.34 | 2.20 | - | - | - | - | - | - | - | - | - | .16 | .27 | .18 | -.18 | -.43* |
| 10. Goal facilitation (time 2) | 5.79 | 2.49 | - | - | - | - | - | - | - | - | - | - | .09 | .10 | -.06 | .17 |
| 11. Anxiety symptoms (time 3) | 3.97 | 2.85 | .74 | - | - | - | - | - | - | - | - | - | - | .54** | -.31 | -.72** |
| 12. Depressive symptoms (time 3) | 1.90 | 1.92 | .61 | - | - | - | - | - | - | - | - | - | - | - | -.53** | -.62** |
| 13. Physical health (time 3) | 87.81 | 16.28 | .91 | - | - | - | - | - | - | - | - | - | - | - | - | .73** |
| 14. Psychosocial health (time 3) | 76.64 | 15.24 | .91 | - | - | - | - | - | - | - | - | - | - | - | - | - |

*Note. *p < .05; **p < .01*

**Analyses**

More acceptance at time 1 was significantly related to less anxiety symptoms at time 3 (after controlling for gender; $c' = -.44$, $SE = .17$, $p = .01$), less depressive symptoms at time 3 (after controlling for health status; $c' = -.33$, $SE = .10$, $p = .00$), but unrelated to psychosocial health (after controlling for health status and negative life events; $c' = 1.53$, $SE = .90$, $p = .10$), and physical health (after controlling for age, time since diagnosis, health status and negative life events; $c' = .66$, $SE = 1.07$, $p = .54$). In each mediation model, acceptance at time 1 was not related to goal interference (range $\alpha = -.26 - -.21$, range $SE = .13 - .15$, range $p = .09 - .14$) or goal facilitation (range $\alpha = -.18 - -.10$, range $SE = .16 - .17$, range $p = .28 - .52$) measured at time 2. Furthermore, there was also no significant relationship between goal interference and facilitation at time 2 and anxiety symptoms ($b = .17$, $SE = .21$, $p = .44$; $b = -.14$, $SE = .19$, $p = .48$), depressive symptoms ($b = -.08$, $SE = .14$, $p = .56$; $b = -.01$, $SE = .12$, $p = .91$), psychosocial health ($b = -1.56$, $SE = 1.22$, $p = .21$; $b = -.37$, $SE = 1.00$, $p = .71$), and physical health ($b = 1.05$, $SE = 1.37$, $p = .45$; $b = -.57$, $SE = 1.20$, $p = .64$) at time 3. Accordingly, there were no significant indirect effects of acceptance at time 1 through goal interference/facilitation at time 2 on anxiety symptoms [$ab_{int} = -.04$, $SE_{int} = .07$, $CI_{int}$: $(-.25, .05)$; $ab_{fac} = .01$, $SE_{fac} = .05$, $CI_{fac}$ $(-.04, .21)$], depressive symptoms [$ab_{int} = .02$, $SE_{int} = .04$, $CI_{int}$: $(-.04, .15)$; $ab_{fac} = .00$, $SE_{fac} = .03$, $CI_{fac}$ $(-.04, .08)$], psychosocial health [$ab_{int} = .33$, $SE_{int} = .53$, $CI_{int}$: $(-.17, 2.15)$; $ab_{fac} = .07$, $SE_{fac} = .28$, $CI_{fac}$ $(-.27, 1.02)$], and physical health [$ab_{int} = -.11$, $SE_{int} = .46$, $CI_{int}$: $(-1.40, .49)$; $ab_{fac} = .06$, $SE_{fac} = .42$, $CI_{fac}$ $(-.50, 1.24)$] at time 3 as their bias corrected (BC) bootstrapped confidence interval (95% BC with 5000 resamples) included zero.

**DISCUSSION**

This study, as a first, longitudinally investigated whether the relationship between acceptance and well-being in adolescents with chronic illness is mediated by interference and facilitation of life goals by treatment goals. Analyses revealed that acceptance predicted certain aspects of well-being (i.e., anxiety and depressive symptoms). However, these relationships were not explained by the interference or facilitation of life goals by treatment goals.

Consistent with previous research (e.g., Casier et al., 2008, 2011), acceptance at time 1 was found to predict particular aspects of well-being at time 3, i.e., anxiety and depressive symptoms. Contrary to this research and our expectations, acceptance did not

relate to other aspects such as quality of life. The generally high levels of quality of life and acceptance in the current sample may explain this lack of findings. The finding that more acceptance relates to less anxiety and depressive symptoms fits the conceptualisation of acceptance as 'a force to engage in what is valued in life despite being ill', and 'a way towards increasing investment in valuable life domains instead of a sign of weakness, resignation, and failure' (Evers et al., 2001; Hayes et al., 2006; Risdon et al., 2003).

For the first time, it was also examined whether the relationship between acceptance and well-being was explained by goal processes such as the experience of interference (when striving for one goal decreases the chance of attaining another goal) or facilitation (when striving for one goal increases the chance of attaining another goal) between goals at time 2. This was expected because acceptance is defined as a means towards engagement in important life goals, despite being ill and because goal processes are assumed to impact well-being in the context of chronic illness (De Ridder & Kuijer, 2007; Evers et al., 2001; Gebhardt, 2008; Hayes, Luoma, Bond, Masuda, Lillis, 2006; Maes & Karoly, 2005; Risdon, Eccleston, Crombez, & McCracken, 2003). Contrary to our expectations, this was not the case. First, acceptance did not relate to goal interference and facilitation. Given the definition of acceptance as 'adapting to chronic illness' and 'handling averse consequences' (Evers et al., 2001, p. 1027), it may be that this conceptualisation of acceptance does not relate to the experience of goal interference and facilitation per se, but to how individuals handle these experiences. Future research should explore this explanation, by investigating if acceptance relates to other goal constructs such as a reconciliation between treatment and other life goals, and the importance and achievability of these goals. Second, goal interference and facilitation at Time 2 did also not relate to well-being at Time 3. This is contrary to previously found relationships between goal interference and well-being but in line with the previously found lack of association between goal facilitation and well-being (for an overview see Riediger, 2007). Several explanations can be proposed for these unexpected results concerning goal interference. It is possible that the time interval between the measurement of goal interference and well-being was not optimal (e.g., too short) to unfold an effect (Cole & Maxwell, 2003). Therefore, future research should investigate this effect for a variety of time intervals. Also, it may be that not goal interference itself, but rather the negative emotions that follow this interference may affect well-being (Massey et al., 2009a; Massey, Garnefski, Gebhardt, & van der Leeden, 2009b).

Moreover, the content of treatment goal of the participating adolescents was predefined as 'doing treatment'. This definition may be too abstract and/or comprehensive. Further research should focus on those aspects of treatment that adolescents themselves identify as very important. Third, there also was no indirect effect of acceptance at Time 1 through goal interference and facilitation at Time 2 on well-being at Time 3.

This study also, as a first, explored the relationship between goal facilitation and well-being in the context of chronic illness. Findings on goal facilitation in healthy individuals is inconsistent as it reports that facilitation was mostly unrelated, but sometimes positively related to well-being (Riediger, 2007; Riediger & Freund, 2004). The current results add to the majority of findings that goal facilitation by treatment goals does not relate to well-being. This lack of association may be explained by the fact that individuals tend to be rather unresponsive to gains (see Riediger, 2007). Comparing experienced goal interference and facilitation between individuals with chronic illness and healthy peers may be an interesting avenue for future research. This research may indicate if it is necessary to further address goal interference and facilitation in chronically ill adolescents, and may point to indications for intervention development.

Some limitations need to be considered when interpreting the current results. First, the response rate was rather low. As participating and non-participating adolescents may significantly differ on certain characteristics (e.g., better health status), this should be taken into account when interpreting the results. Second, the current sample seems to consist of well-functioning adolescents considering the average high levels of acceptance and goal facilitation and low levels of goal interference. Therefore, these results may not be transferable to less well-functioning adolescents. Third, our measure of acceptance does not fully capture the richness of the construct of acceptance as it has been developed in scientific and clinical use. New measures need to be developed that, next to assessing how to deal with the nature and consequences of chronic illness, also address other core aspects of acceptance such as goal engagement and the embrace of unwanted events (Casier et al., 2011; Hayes et al., 2004). This measure could be based on the Acceptance and Action Questionnaire (AAQ; Hayes et al., 2004), a measure that has already been adapted for diabetes but not yet for CF.

The current findings have important implications for intervention in adolescents with chronic illness. Given the relationship between acceptance and anxiety and depression, psychological interventions promoting acceptance may be particularly helpful for adolescents with chronic illness. Acceptance-based interventions, such as Acceptance

and Commitment Therapy (ACT; Hayes, 2004) are already available for paediatric sickle cell disease and chronic pain and seem to lead to less symptoms, and an improved functioning and quality of life (e.g., Masuda et al., 2011; Wicksell et al., 2009). Further research replicating the current findings is needed to find out whether these interventions are useful for adolescents with CF and diabetes. If the well-being of these adolescents indeed can be increased and protected by acceptance, existing interventions such as ACT (Hayes, 2004) need to be developed for and tested in the context of CF and diabetes.

### REFERENCES

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Caes, L., Vervoort, T., Eccleston C., & Goubert, L. (2012). Parents who catastrophize about their child's pain prioritize attempts to control pain. *Pain, 153,* 1695-1701.

Casier, A., Goubert, L., Huse, D., Theunis, M., Franckx, H., Robberecht, E., Matthijs, D., & Crombez, G. (2008). The role of acceptance in psychological functioning in adolescents with cystic fibrosis: a preliminary study. *Psychology and Health, 23(5),* 629-638. doi: 10.1080/08870440802040269

Casier, A., Goubert, L., Theunis, M., Huse, D., De Baets, F., Matthys, D., & Crombez, G. (2011). Acceptance and well-being in adolescents and young adults with cystic fibrosis: a prospective study. *Journal of Pediatric Psychology, 36,* 476-487. doi: 10.1093/jpepsy/jsq111

Cella, D. (1998). Quality of life. In J.C. Holland (Ed.), Psychooncology (pp. 1135-1146). New York, Oxford: Oxford University Press.

Diener, E. (1984). Subjective well-being. *Psychological Bulletin, 95(3),* 542-575.

Diener, E., Suh, E.M., Lucas, R.E., & Smith, H.L. (1999). Subjective well-being: Three decades of progress. *Psychological Bulletin, 125,* 276-302.

Evers, A.W.M., Kraaimaat, F.W., van Langenveld, W., Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036.

Gjengedal, E., Rustøen, T., Wahl, A.K., & Hanestadt, B.R. (2003). Growing up and living with cystic fibrosis. Everyday life and encounters with the health care and social services – A qualitative study. *Advances in Nursing Science, 26 (2),* 149-159.

Glasscoe, C. A., & Quittner, A. L. Psychological interventions for people with cystic fibrosis and their families. Cochrane Database of Systematic Reviews 2008, Issue 3. Art. No.: CD003148. doi: 10.1002/14651858.CD003148.pub2.

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: model, processes and outcomes. *Behaviour Research and Therapy, 44,* 1-25. doi:10.1016/j.brat.2005.06.006

Little, B. (1983). Personal Projects Analysis. Retrieved from http://www.brianrlittle.com/Topics/research/assessment-tools/

Little, B. R. (1983). Personal projects: A rationale and method for investigation. *Environment and Behavior, 15,* 273–309.

Maes, S. & Karoly, P. (2005). Self-regulation assessment and intervention in physical health and illness: a review. *Applied Psychology: An International Review, 54(2),* 267-299.

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: Acceptance and Commitment Therapy for pediatric sickle cell disease. *Journal of Pediatric Psychology, 36,* 398-408. doi:10.1093/jpepsy/jsq118

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,* 421-460. doi: 10.1016/j.dr.2008.03.002

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents and young adults with chronic pain: validation of an adapted assessment instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi:10.1016/j.ejpain.2009.05.002

Nurmi, J.E. (1987). Age, sex, social class, and quality of family interactions as determinants of adolescents' future orientation: A developmental task interpretation. *Adolescence, 22,* 977–991.

Nurmi, J.E. (1991). How do adolescents see their future? A review of the development of future orientation and planning. *Developmental Review, 11,* 1–59.

Nurmi, J.E. (1993). Adolescent development in an age-graded context: The role of personal beliefs, goals, and strategies in the tackling of developmental tasks and standards. *International Journal of Behavioral Development, 16,* 169–189.

Pinquart, M., & Teubert, D. (2012). Academic, physical, and social functioning o children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 37,* 376-389. doi: 10.1093/jpepsy/jsr106

Pinquart, M., & Shen, Y. (2011). Behavior problems in children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 36,* 1003-1016. doi: 10.1093/jpepsy/jsr042

Riediger, M. (2001). On the dynamic relations among multiple goals: Intergoal conflict and intergoal facilitation in younger and older adulthood. (Doctoral dissertation, Free University, Berlin, Germany). Retrieved from http://www.diss.fu-berlin.de/2001/266/

Riediger, M. (2007). Interference and facilitation among personal goals: Age-group differences and associations with well-being and behavior. In B.R. Little, K. Salmela-Aro, J.-E. Nurmi, & S.D. Philipps (Eds.), Personal project pursuit: Goals action, and human flourishing (pp. 119-143). Erlbaum.

Riediger, M. & Freund, A.M. (2004). Interference and facilitation among personal goals: differential associations with subjective well-being and persistent goal pursuit. *Personality and Social Psychology Bulletin, 30,* 1511-1523. doi: 10.1177/0146167204271184

Risdon, A., Eccleston, C., Crombez, G., & McCracken, L. (2003). How can we learn to live with the pain? A Q-methodological analysis of the diverse understandings of acceptance of chronic pain. *Social Science and Medicine, 56,* 375-386. doi: 10.1016/S0277-9536(02)00043-6

Schwartz, L. A., & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: A review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 390–405. doi:10.1007/s10880-006-9041-8

Schwartz, L. A., & Drotar, D. (2009). Health-related hindrance of personal coal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and

peers without a history of chronic illness. *Journal of Pediatric Psychology, 39,* 954–965. doi:10.1093/jpepsy/jsn144

Seiffge-Krenke, I. (1998). Chronic disease and perceived developmental progression in adolescence. *Developmental Psychology, 34,* 1073–1084. doi:10.1037/0012-1649.34.5.1073

Seiffge-Krenke, I. (2001). Diabetic adolescents and their families: stress, coping, and adaptation. Cambridge, UK: Cambridge University Press.

Spilker (1996). Introduction. In B. Spilker (Ed.), Quality of life and pharmacoeconomics in clinical trials (pp. 1-10). Philadelphia, PA: Lippincott-Raven.

Vervoort, T., Huguet, A., Verhoeven, K., & Goubert, L. (2011). Mothers' and fathers' responses to their child's pain catastrophizing and disability. *Pain, 152,* 786-793. doi: 10.1016/j.pain.2010.12.010

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi:10.1016/j.pain.2011.09.003

Yeo, M. & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330,* 721-723. doi: 10.1136/bmj.330.7493.721

# GENERAL DISCUSSION

## PREFACE

In adolescents with chronic illness, the permanent health threat and demanding treatment may seriously impact upon well-being. More specifically, growing up with chronic illness is a risk factor for several difficulties such as depressive symptoms, behaviour problems, and impairment in academic, physical, and social functioning (see Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Schwartz & Drotar, 2006, 2009; Yeo & Sawyer, 2005). Therefore, it is essential to understand how adolescents with chronic illness can maintain their well-being despite the burden of being ill.

Within this dissertation, 'acceptance of illness' was identified as a potential protective factor for the well-being of adolescents with chronic illness (Evers et al., 2001). Indeed, previous research in adolescents with chronic illness has indicated that acceptance may play a protective role in the well-being of children and adolescents with chronic illnesses such as chronic pain (e.g., McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011), juvenile arthritis (Feinstein et al., 2011), and sickle cell disease (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011). Specifically, this research has shown that more acceptance is related to positive outcomes such as less anxiety, less depression, less disability, and better emotional, social, and physical functioning.

Next to investigating the role of acceptance of illness in well-being, understanding of the processes through which acceptance may affect well-being is essential. The current dissertation focussed on two specific goal regulation processes (i.e., goal interference and goal facilitation) as possible mediators. These goal regulation processes are especially important for adolescents with chronic illness as during adolescence individuals need to achieve important life tasks to pass on to adulthood, start to regulate their goals more autonomously, are cognitively capable to evaluate the impact of illness on goal regulation, have a strong desire to be 'normal', tend to be non-adherent to treatment, and need to pursue treatment goals which may interfere with the pursuit of other life goals (e.g., becoming independent) (Badlan, 2006; Little, 1983; Massey, Gebhardt, & Garnefski, 2008; Nurmi, 1987, 1991, 1993; Schwartz & Drotar, 2006, 2009).

Central aims of the current dissertation were to investigate (1) the role of acceptance of illness in the well-being of adolescents with chronic illness, (2) the role of acceptance in the experienced goal interference and facilitation of these adolescents, (3) the mediating role of goal interference and facilitation in the relationship between acceptance and well-being, and (4) the differences in goal regulation between adolescents with chronic illness and their healthy peers. These aims were investigated in six studies in adolescents with cystic fibrosis (CF) or diabetes and healthy peers, by means of different research methods.

This discussion will first provide an overview of the main research findings. Also theoretical and clinical implications, and limitations of the current set of studies will be discussed. Finally, possible avenues for future research will be highlighted.

## MAIN FINDINGS

**Chapter 1** reported on the findings of a cross-sectional questionnaire study focussing on the relationship between acceptance and a first component of well-being: negative affect. The sample of this study consisted of 34 adolescents with CF between 12 and 22 years. All adolescents completed a battery of self-report questionnaires assessing acceptance, anxiety, depressive symptoms, and functional disability. In line with expectations, results suggested that more acceptance of illness was related to less anxiety, less depressive symptoms, and less functional disability beyond the effects of disease severity. **Chapter 2** aimed to replicate these findings but also extended them by using a prospective design and by also focussing on the relationship between acceptance and a second component of well-being, i.e. life satisfaction. This prospective questionnaire study was conducted in 40 adolescents with CF (14-22 years). Acceptance, anxiety, depression, and physical, emotional, role, and social functioning were assessed by self-report questionnaires. These questionnaires were completed at baseline (Time 1) and six months later (Time 2). Again it was found that more acceptance was related to less depressive symptoms (at both Time 1 and 2) but, contrary to expectations, not to anxiety. Furthermore findings revealed that more acceptance was related to a better emotional, role, and social functioning at Time 1 but not Time 2. Acceptance, however, was unrelated to physical functioning. In **chapter 3** we again investigated the relationship between acceptance and negative affect. Different from chapter 1 and 2, negative affect was measured during a real-life situation. Thirty-six adolescents with CF (12-22 years) completed a questionnaire assessing acceptance of illness and reported on the amount of

expected and experienced pain and distress during a medical procedure they had to undergo in the hospital (i.e., spirometry). Also physiological measures of distress were recorded (i.e., heart rate and heart rate variability). Findings revealed that more acceptance was related to less expected pain and pain-related thoughts, but not to distress prior to and during spirometry, nor to experienced pain. In **chapter 4**, goal regulation was introduced. Goals and goal regulation processes were compared in a questionnaire study in adolescents with ($N = 60$; 14-22 years) and without chronic illness ($N = 60$; 14-22 years). It was found that adolescents with chronic illness rated their health goals as more important and more difficult to achieve than healthy peers but, surprisingly, reported to be more successful in attaining their health goal. Also, adolescents with and without chronic illness did not differ on the reported importance, success, and difficulty of their non-health goals, except for the difficulty in attaining their relationship goal, which was perceived as less difficult in adolescents with chronic illness. Finally, both groups did not differ on the amount of experienced interference of non-health goals by health goals. **Chapter 5** proceeded on investigating goal regulation processes by determining whether daily experienced interference and facilitation of life goals by treatment goals in adolescents with CF and diabetes mediated the relationship between acceptance and well-being. This study was conducted in 38 adolescents with CF or diabetes (14-22 years) and made use of a daily process design. Next to assessing acceptance, negative life events, and goal-related self-efficacy by means of questionnaires, daily mood, daily experienced interference and facilitation of life goals by treatment goals were assessed by an online diary completed during three consecutive weeks. Findings revealed that acceptance of illness was positively related to daily well-being, but unrelated to daily goal interference and facilitation. Furthermore, daily goal interference and facilitation were unrelated to same and next day well-being. In **chapter 6** we aimed to replicate the findings from chapter 5, using a different methodology. This time, a longitudinal instead of a daily process design was used. Thirty participants with CF or diabetes completed questionnaires assessing acceptance, negative life events, goal-related self-efficacy, well-being, and interference and facilitation of life goals by treatment goals at three points in time (baseline, two, and four months later). Again it was found that acceptance of illness (at Time 1) was positively related to some dimensions of well-being (at Time 3; i.e., anxiety and depressive symptoms, but not health-related quality of life), but unrelated to goal interference and facilitation (at Time 2). Furthermore, goal interference and

facilitation (at Time 2) were unrelated to all aspects of well-being (at Time 3). Chapter 6 thus confirmed the findings from chapter 5.


## THEORETICAL IMPLICATIONS

### The role of acceptance of illness in the well-being of adolescents with CF or diabetes

A first aim of this dissertation was to investigate the relationship between acceptance of illness and well-being in chronic illness. The findings of the current set of studies contributed to the increasing support that acceptance indeed relates to well-being in the context of chronic illness, beyond the effects of socio-demographic and disease-related variables. Specifically, chapter 1, 2, 3, 5, and 6 of this dissertation indicated that higher acceptance of illness relates to several aspects of well-being such as less anxiety, less depressive symptoms, less functional disability, better emotional, role, and social functioning, less expected pain and pain-related thoughts prior to medical procedures, and higher positive and lower negative daily mood. These findings were in line with recent research in adults and adolescents with chronic pain (e.g., McCracken et al., 2010; McCracken & Vowles, 2008; McCracken & Zhao-O'Brien, 2010; Wicksell et al., 2011), juvenile arthritis (Feinstein et al., 2011), chronic fatigue syndrome (Van Damme, Crombez, Van Houdenhove, Mariman, & Michielsen, 2006), tinnitus (Westin, Hayes, & Andersson, 2008), multiple sclerosis (Evers et al., 2001; Pakenham, 2006), and sickle cell disease (Masuda et al., 2011) indicating that higher levels of acceptance relate to less pain, less anxiety and depression, better functioning, better (health-related) quality of life, better mood, less physical complaints, less distress, and less disability. The current findings also extended this research by suggesting that the relationship between acceptance and well-being also occurs in adolescents with other chronic illnesses such as CF and diabetes.  Furthermore, the findings of chapter 3 carefully suggested that acceptance may relate, not only to chronic, but also acute consequences of illness such as expected pain and pain-related thoughts prior to medical procedures (i.e., spirometry). Based on the current findings it may be concluded that acceptance relates to the adjustment to living with chronic illness in general (see chapter 1, 2, 5, and 6), but also to very specific illness-related experiences such as expected pain during spirometry and pain-related thoughts (see chapter 3).

During adolescence, chronic illness and its burden may become more severe, specific disease patterns and symptoms may emerge, and more intensive medical follow up may be required. At the same time, a growing personal responsibility for treatment

needs to be developed (Ernst, Johnson, & Stark, 2010; Michaud, Suris, & Viner, 2004). Next to managing these challenges, it is important for affected adolescents to maintain and sustain their well-being. This means that a delicate balance between disease-related challenges and developmental challenges becomes essential during this period (e.g., doing treatment, getting hospitalised versus building social relationships, getting an academic degree, becoming independent; Badlan, 2006; Gjengedal, Rustøen, Wahl, & Hanestad, 2003; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009). Acceptance may impact this balance as it may help adolescents with chronic illness to keep pursuing important developmental goals (e.g., independence, close relationships, academic achievement, family life, occupation) in the development towards adulthood (Ernst et al., 2010; Gjengedal et al., 2003). Examples of this balance may be the adjustment of developmental goals that have become unrealistic because of illness (e.g., having a part-time job instead of a full-time, postpone university studies and first get health back on track) or the integration of disease-related goals and important developmental goals (e.g., doing aerosol therapy while studying).

Despite the use of prospective, daily process, and longitudinal designs (see chapter 2, 5, and 6), no univocal conclusions about the direction of the relationship between acceptance and well-being can be drawn. Chapter 2 provided preliminary support for acceptance as a predictor of depressive symptoms, but not of anxiety and physical, emotional, role, and social functioning. Chapter 6 also suggested that acceptance may serve as a predictor of depressive symptoms and anxiety, but also not for physical, emotional, role, and social functioning. Chapter 5, on the other hand, provided no support for acceptance as a predictor of daily positive and negative mood. This lack of clarity concerning the causal status of acceptance of illness may likely be due to the lack of power of these studies, whereby smaller effects remained undetected. Further research, using larger samples, is needed to clarify this issue.

**The role of acceptance in the experienced goal interference and facilitation in adolescents with CF or diabetes**

A second aim of the current study was to examine the relationship between experienced goal interference and facilitation in adolescents with CF or diabetes. The studies described in chapter 5 and 6 of this dissertation were, to our knowledge, the first to investigate this relationship. Specifically, the relationship between acceptance and experienced goal interference and facilitation of life goals by treatment goals was investigated. Results revealed that, contrary to expectations, acceptance was not related to

goal interference, nor to goal facilitation. Possible explanation for this lack of association is that acceptance of illness rather relates to the way one deals with the experiences of goal interference and facilitation instead of to the experience of goal interference and facilitation per se. Furthermore, acceptance may rather be associated with other goal-related constructs such as a balance between treatment goals and other important life goals, the relative importance of these goals, and the amount of success in the pursuit of these goals. Acceptance indeed comprises engaging in important life goals despite being ill (Evers et al., 2001; Hayes, Luoma, Bond, Masuda, & Lillis, 2006). This may mean that a balanced interplay between treatment goals (e.g., doing treatment) and other important life goals (e.g., building social relationships, getting an academic degree, becoming independent) is essential. Further research is needed to determine whether acceptance indeed relates to these constructs and experiences.

**The mediating role of goal interference and facilitation in the relationship between acceptance and well-being in adolescents with CF or diabetes**

A third aim of this dissertation was to determine whether the relationship between acceptance and well-being in adolescents with CF or diabetes is mediated by goal interference and facilitation. Insight in the role of goal interference or facilitation is essential during adolescence because striving for  important life goals in the transition to adulthood may be more difficult for adolescents with chronic illness than for healthy peers (Badlan, 2006; Schwartz & Drotar, 2006). As already discussed above, acceptance was related to well-being, but not related to goal interference and facilitation. Furthermore, the results from chapter 5 and 6 further revealed that the previously established relationship between goal interference and well-being in healthy individuals (for an overview see Riediger, 2007) has not been confirmed in the context of chronic illness. Specifically, goal interference of life goals by treatment goals was unrelated to (daily) well-being. Furthermore, it was found that, in chronic illness, goal facilitation of life goals by treatment goals was unrelated to well-being, which is in line with the majority of research in healthy individuals (for an overview see Riediger, 2007). These findings indicated that the degree to which the pursuit of treatment goals makes the pursuit of other valued goals more difficult or easier does not relate to the degree of well-being in adolescents with CF and diabetes. It can thus be concluded that the relationship between acceptance and well-being in adolescents with CF or diabetes was not mediated by goal interference and facilitation.

Several explanations can be proposed for the unexpected results regarding goal interference. It is possible that the time between the assessment of goal interference and the assessment of well-being was not optimal (e.g., too short) for an effect to unfold (Cole & Maxwell, 2003). Therefore, future research should investigate this relationship within different time frames. It may also be that not goal interference itself, but rather the emotional reactions resulting from this interference may affect well-being. This explanation is in line with recent research suggesting that not the extent to which goals are blocked but the frustration that occurs upon it relates to well-being (Massey, Gebhardt, & Garnefski, 2009a; Massey, Garnefski, Gebhardt, & van der Leeden, 2009b). Further research is needed to investigate this hypothesis. Goal interference may be constructively handled by several strategies such as searching for compatible strategies to attain different goals, identifying instrumental relations between goals, optimising the use of limited resources (e.g., time, energy), searching for other goals in the same domain or for alternative routes to goals, scaling back goals, or even disengaging from incompatible goals and reengaging in other compatible goals (e.g., choosing active instead of sedentary leisure activities that match with treatment guidelines regarding physical activity). The lack of association between goal interference and well-being may also be due to methodological issues such as difficulties with understanding the instructions concerning the ratings of goal interference. Furthermore, the lack of association between goal facilitation and well-being may be explained by the fact that individuals generally do not react strongly to gains (see Riediger, 2007).

**Differences in goal regulation between adolescents with CF or diabetes and their healthy peers**

A final aim of the current dissertation was to compare goal setting and striving between adolescents with and without CF or diabetes. The study described in chapter 4 of this dissertation compared adolescents with and without CF or diabetes on those aspects of goal setting and striving that are assumed to be affected by chronic illness (i.e., new health goals, reprioritization and adaptation of goals, and changes and/or hindrance in goal striving). Specifically, this study compared these adolescents on the content of their health goals, the importance of and the success, and difficulty in attaining their (non-)health goals, and the amount of experienced interference between their health and non-health goals. The findings of this study showed that the content of the most important health goals reported by adolescents with chronic exclusively related to their illness. This is in line with previous findings suggesting that chronic illness forces to set new goals in

response to this illness (e.g., 'taking medicines', 'adhering to treatment'; see Schwartz & Drotar, 2006 for a review). Furthermore, adolescents with chronic illness rated their health goals as more important and more difficult to achieve than healthy peers, but reported to be more successful in attaining their health goals. These findings concerning difficulty, but not regarding success, are in line with previous research showing that chronic illness causes changes and/or hindrance in goal striving (e.g., having difficulties to strive for goals because of physical complaints or limitations, showing less commitment to goals), but can also be credited to the complex and demanding character of health goals (Badlan, 2006; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2006, 2009; Seiffge-Krenke, 2001). The seeming contradiction between the findings concerning success and difficulty may be explained by a high sense of self-efficacy that adolescents with chronic illness may have concerning their health goals, or by a stronger commitment to their health goals, or by the possession of certain self-regulatory skills (i.e., planning, self-monitoring), whereby more difficulty does not necessarily translates into less success (Maes & Karoly, 2005). In contrast to the results regarding health goals, both groups rated the importance, success, and difficulty of their non-health goals similarly, except for the difficulty in attaining their relationship goal, which was perceived as less difficult in the adolescents with chronic illness. The findings regarding importance revealed that adolescents with CF or diabetes give the same priority to their non-health goals as their healthy peers, what may suggest that adolescents with chronic illness are able to adjust and reconcile both their health and non-health goals (see Schwartz & Drotar, 2006 for a review). Ways to tune different goals to each other without changing their priority, may be the use of compatible strategies to attain different goals (e.g., 'join a running team' may be effective for both 'obtain a better lung functioning' and 'meet new people'), the identification of possible instrumental relations between goals (e.g., 'take responsibility for treatment' may be a step towards 'become independent'), and the optimization of the use of limited resources (e.g., time, energy; e.g., 'doing aerosol treatment while studying') (Riediger & Freund, 2004). Finally, both groups reported an equal amount of experienced interference of non-health goals by health goals. This was not in line with previous research suggesting that chronic illness may lead to hindrance in goal striving (see Schwartz & Drotar, 2006 for a review). Yet, it suggested that the chronically ill adolescents in this study know how to balance the demands of chronic illness with their regular life goals, without disengaging from or scaling back on these goals (cfr. equal ratings on goal importance). Such balance may be realized by, on top of the strategies

mentioned above, a focus on more realistic goals or an adjustment of certain goals to situational constraints (e.g., part-time instead of full-time education) (Brandstätter & Renner, 1990; Schwartz & Drotar, 2009). Future research should investigate which strategies adolescents and young adults with chronic illness exactly use to adapt and reconcile their goals.

This study showed that goal setting and striving is largely similar in the adolescents with chronic illness and their healthy peers from the current study, except for some aspects of their health goals. Based on these results, it may be concluded that some adolescents with CF or diabetes seem well-adapted to their illness, seem able to balance their health and other life goals, and seem to feel confident in striving for their health goals. The current study extended previous research comparing goal setting and striving in adolescents with diabetes or cancer with that in healthy peers (Helgeson & Takeda, 2009; Schwartz & Parisi, in press; Seiffge-Krenke, 1998). These previous studies solely focused on the content and number of identified life goals, possible delay in these goals, and the achievability of these goals. They found that adolescents with diabetes did not differ in the number of identified life goals from healthy peers, but that adolescents with cancer identified fewer goals. Furthermore, these studies found that adolescents with diabetes identified less self-improvement and more appearance goals, whereas adolescents with cancer were less likely to identify interpersonal or leisure goals compared with their healthy peers. Finally, findings from these studies revealed that adolescents with diabetes perceived delays in some of their developmental goals (i.e., physical maturity and individual lifestyle) and that adolescents with cancer rated their goals as more achievable as compared with healthy peers (Helgeson & Takeda, 2009; Schwartz & Parisi, in press; Seiffge-Krenke, 1998). Chapter 4 of this dissertation added to this research by focussing on those specific aspects of goal setting and striving that are assumed to be affected by chronic illness (i.e., new health goals, reprioritization and adaptation of goals, and changes and/or hindrance in goal striving).

**Conceptualisation of acceptance**

In this dissertation acceptance was defined as 'recognizing the need to adapt to chronic illness while perceiving the ability to tolerate the unpredictable, uncontrollable nature of the disease and handle its averse consequences' (Evers et al., 2001, p.1027). Acceptance refers to acknowledging the reality of being ill, while perceiving oneself as able to live with the consequences of illness (Evers et al., 2001). This conceptualisation suggests that the ability to reconcile to the limitations a chronic illness involves, and to

face the variable demands imposed by this illness while living a valuable life are central in acceptance (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003).

Several findings from the studies described in this dissertation provide support for this conceptualisation. First, the significant negative relationship between acceptance and anxiety and depressive symptoms described in chapter 1 and 2 of this dissertation were in line with this conceptualisation. Acknowledging the reality of being chronically ill, while believing one can live with illness and that illness does not imply the end of a meaningful life seems indeed inconsistent with anxiety and depressive symptoms (Evers et al., 2001; Hayes et al., 2006; Risdon, Eccleston, Crombez, & McCracken, 2003). Second, in chapter 2, 5, and 6 it was also found that more acceptance relates to a better quality of life and daily well-being, which was also in line with the conceptualisation of acceptance. Acceptance indeed includes the perception to be able to live with the consequences of illness, while living a valuable life despite being ill (Evers et al., 2001; Hayes et al., 2006; Risdon et al., 2003). Third, the findings described in chapter 3 of this dissertation, cautiously indicated that acceptance of illness may also comprise the ability to face more indirect demands of having a chronic illness, such as the need to undergo certain medical procedures. This suggested that acceptance of illness not only concerns adjustment to illness in general, but also the adjustment to very specific illness-related experiences such as undergoing medical procedures. Fourth, the unexpected lack of association between acceptance of illness and the experience of goal interference and facilitation described in chapter 5 and 6 also added to the understanding of acceptance. These findings may indicate that acceptance rather refers to the way of dealing with goal interference and facilitation, than with the experience of interference and facilitation itself. Acceptance is indeed defined in terms of 'adapting to chronic illness' and 'handling averse consequences' (Evers et al., 2001, p. 1027). Therefore, it may be the case that acceptance rather relates to other goal constructs such as a balance between treatment goals and other important life goals, the relative importance of these goals, and the amount of success in the pursuit of these goals. Acceptance indeed includes engaging in a valuable life despite being ill (Evers et al., 2001; Hayes et al., 2006). To obtain a valued life, a delicate balance between treatment goals (e.g., doing treatment) and other important life goals (e.g., building social relationships, getting an academic degree, becoming independent) may be essential. Future research should investigate if this is the case.

**CLINICAL IMPLICATIONS**

The findings from this dissertation concerning acceptance (see chapter 1, 2, 3, 5, and 6) have several implications for clinical practice. As acceptance relates to several aspects of well-being, adolescents with chronic illness may benefit from psychological interventions that promote acceptance. These interventions may sustain well-being and/or enhance well-being when affected. Interventions focussing on acceptance, such as Acceptance and Commitment Therapy (ACT; Hayes, 2004) are already developed for children and adolescents with sickle cell disease and chronic pain and lead to less symptoms, an improved functioning, and a better quality of life (e.g., Masuda et al., 2011; Wicksell, Melin, Lekander, & Olsson, 2009). Within ACT (Hayes, 2004; Hayes et al., 2006), acceptance is defined as a means to increase action toward important life goals, in spite of being ill (Bach & Hayes, 2002; Evers et al., 2001; Hayes et al., 2006). This may be essential during adolescence as adolescents with chronic illness need to obtain a balance between health goals (i.e., disease management) and other important life goals (e.g., independence, close relationships, academic achievement, family life, occupation) (Badlan, 2006; Gjengedal et al., 2003; Glasscoe & Quittner, 2008; Schwartz & Drotar, 2009). By acknowledging the reality of being ill, by being confident to be able to live with the consequences of illness, and by believing that chronic illness is not the end of a meaningful life and, a meaningful life and well-being can be sustained (Evers et al., 2001; Hayes et al., 2006; Risdon et al., 2003). Additional research is needed to verify whether ACT is useful for adolescents with CF and diabetes.

The comparison of goal setting and striving between adolescents with and without CF or diabetes revealed that goal setting and striving is largely similar in both groups, except for some aspects of their health goals (see chapter 4). Despite the preponderance of similarities, it is important for clinical practice to accurately monitor the goal setting and striving of adolescents with CF or diabetes and to act appropriately when difficulties occur. Examples of such difficulties can be the experience of life goal interference by health goals, the unnecessary disengagement from valued and essential life goals, an unbalance in health and non-health goals, etc.

As the findings from this dissertation concerning goal interference and facilitation revealed no significant results (see chapter 5 and 6), no recommendations for clinical practice regarding these processes can be made. However, as the samples from the studies described in chapter 5 and 6 were rather small and consisted of adolescents reporting low levels of interference, these studies provide no evidence for the actual

absence of an effect between acceptance and interference/facilitation, and between interference/facilitation and wellbeing. Therefore, further research investigating these relationships is needed.

## LIMITATIONS

Some limitations need to be considered when interpreting the findings from this dissertation. First, the studies discussed in chapter 1, 2, 3, 5, and 6 only focused on acceptance in adolescents with CF or diabetes. Consequently, it is not certain whether these results generalize to adolescents with other chronic conditions.

Second, the samples of adolescents with CF or diabetes used in our studies reported on average a good overall functioning (i.e., high levels of acceptance, low levels of disability and psychological difficulty, low levels of distress and pain, high levels of quality of life, and low levels of goal interference). Furthermore, the response rate in most of our studies was rather low, which may reflect a sample bias. Therefore, it cannot be assumed that the current findings apply to the general population of adolescents with CF or diabetes, and in particular to adolescents who function less well.

Third, the studies described in this dissertation mainly made use of self-report measures administered to the adolescents. No assessments from additional informants, such as parents or caregivers, were incorporated to obtain extra information on the functioning and experiences of these adolescents.

Fourth, the sample sizes of all studies presented in this dissertation were rather small. As a result, these studies only had sufficient power to detect large effects. It is therefore possible that small and medium effects remained undetected.

Fifth, several findings (see chapter 1, 2, and 3) were based on cross-sectional and correlational data, and hence, do not indicate causal effects. The prospective study described in chapter 2 is an advancement on these cross-sectional findings, but only provided a first indication of the long-term effects of acceptance. The daily process and longitudinal studies described in chapter 5 and 6 extended these findings by indicating that acceptance may have long-term effects on some, but not all, aspects of well-being studied in this dissertation.

Finally, our conceptualisation of acceptance does not comprise the full richness of the construct of acceptance as developed in scientific and clinical practice. Specifically, the measure of acceptance used throughout this dissertation, lacks the focus on certain facets of acceptance such as dealing with difficult illness-related thoughts and

feelings, engaging in activities that serve important life goals despite being ill, releasing the struggle for change or control, etc. Therefore, new measures that are usable in individuals with CF or chronic conditions in general should be developed. The Acceptance and Action Questionnaire (AAQ) (Badlan, 2006; Evers et al., 2001; Gjengedal et al., 2003; Hayes et al., 2006) could be particularly informative in this regard. The AAQ is a generic measure that assesses ACT processes such as acceptance, values-based action, and psychological flexibility. The AAQ has already been adjusted for individuals with specific conditions such as adolescents and adults with chronic pain (Chronic Pain Acceptance Questionnaire) and adults with diabetes (Acceptance and Action Diabetes Questionnaire; Hayes et al., 2006), but not yet for adolescents with CF.

**FUTURE RESEARCH**

**The role of acceptance of illness in the well-being of adolescents with chronic illness**

Further research is needed to determine whether the positive relationship between acceptance of illness and well-being also occurs in adolescents with other chronic conditions. Until now, the presence of this relationship has been found in adolescents with chronic pain (e.g., McCracken et al., 2010; Wicksell et al., 2011), juvenile arthritis (Feinstein et al., 2011), sickle cell disease (Masuda et al., 2011), and CF and diabetes (see this dissertation). Nonetheless, acceptance may also serve other populations such as adolescents with cancer, tinnitus, asthma, and epilepsy. Furthermore, it is necessary to investigate whether the relationship between acceptance and well-being also occurs in adolescents reporting rather low levels of acceptance and low levels of well-being. Future studies should therefore try to conduct research in more diverse samples, and not only in well-functioning adolescents, which is predominantly the case in the studies reported in the current dissertation.

Future research should also study the long-term effects of acceptance, by using longitudinal designs. This kind of research provides the only way to determine the causal status of acceptance. If it appears that acceptance has a protective role in the well-being of these adolescents, interventions promoting acceptance, such as ACT (Hayes et al., 2006) should be developed for each population studied.

ACT for adolescents has only recently been developed, and has already been used in the context of anorexia (Heffner, Sperry, Eifert, & Detweiler, 2004), chronic pain (e.g., Wicksell, Dahl, Magnusson, & Olsson, 2005), and sickle cell disease (Masuda et al., 2011). These ACT approaches can serve as a basis for the development of ACT for

adolescents with other chronic conditions. By tailoring these approaches to the needs of adolescents with a specific condition, each condition can be met. A great challenge in adapting ACT for adolescents is to make the concepts and exercises used sufficiently clear and comprehensible (Masuda et al., 2011). A more concrete discussion of these concepts and greater focus on behavioural activation may be useful in tackling this challenge (e.g., Wicksell et al., 2005). After these modifications? feasibility studies, starting with case studies or studies in small samples, may be conducted. Finally, randomized controlled trials may be performed to test the efficacy of each ACT approach (Gauthier et al., 2009; Glasscoe & Quittner, 2008). Finally, the mechanism(s) of change in ACT should be investigated. This can be done by conducting mediator analyses (Wicksell et al., 2009, 2011). A possible mechanism of change in ACT may be psychological flexibility, defined as one's ability to fully connect with the present moment, as a conscious human being, and to change or persist in behaviour that is in line with identified values (Hayes et al., 1999). In order to investigate these potential mechanisms, also questionnaires assessing these concepts in adolescents need to be developed (e.g., Ciarrochi, Bilich, & Godsell, 2010; Wicksell et al., 2011).

**The role of acceptance in experienced goal interference and facilitation in adolescents with CF or diabetes**

Future research should further investigate the relationship between acceptance and goal interference and facilitation in the context of chronic illness. As the lack of association in this dissertation may be due to a lack of power, this research should make use of larger samples, so that also small and medium effects can be detected. By conducting such research it can really be determined whether or not an absence of association is the case.

Furthermore, research in adolescents displaying lower levels of acceptance and experiencing more goal interference and less goal facilitation is needed. As the adolescents that participated in the studies from this dissertation may not be representative for the full population of adolescents with CF or diabetes, research in more diverse samples is essential. Otherwise, it cannot be determined whether the findings regarding acceptance and interference and facilitation generalize to all adolescents affected by CF or diabetes.

Besides further investigating the relationship between acceptance and interference and facilitation, the relationship between acceptance and the way adolescents deal with the experience of interference and facilitation may be investigated. This

suggestion fits with the conceptualisation of acceptance described as 'adapting to chronic illness' and 'handling averse consequences' (Evers et al., 2001, p. 1027). It is thus possible that acceptance is rather associated with other goal-related constructs such as a balance between treatment goals and other important life goals, the relative importance of these goals, and the level of success in setting and pursuing these goals. Acceptance indeed includes an engagement in important life goals despite being ill (Evers et al., 2001; Hayes et al., 2006). This may mean that a balanced interplay between treatment goals (e.g., doing treatment) and other important life goals (e.g., building social relationships, getting an academic degree, becoming independent) is essential. Future research should investigate if acceptance indeed relates to these constructs and experiences.

**The mediating effect of goal interference and facilitation on the relationship between acceptance and well-being**

As already mentioned above, future research should investigate whether the relationship between acceptance and well-being is mediated by the way adolescents deal with the experience of interference and facilitation rather than by the experience of interference and facilitation per se. Furthermore, it should examine whether other goal-related constructs such as a balance between treatment goals and other important life goals, the relative importance of these goals, and the amount of success in the pursuit of these goals serve as mediating mechanisms underlying this relationship.

Also, the relationship between goal interference and well-being in the context of chronic illness has to be further explored. This research should make use of  different time intervals between the measurement of goal interference and well-being to verify whether and if so, over what time, the effect of interference unfolds (Cole & Maxwell, 2003). Furthermore, this research should focus, not only on the experience of goal interference itself, but also on the frustration arising from this interference. Indeed, recent research in adolescents has suggested that not the actual number of blocked goals but the frustration resulting from it relates to well-being (Massey et al., 2009a; Massey et al., 2009b).

Finally, research focussing on the impact of treatment goals on other life goals should focus on those aspects of treatment that adolescents themselves identify as very important. In this dissertation the content of treatment goal of the participating adolescents was fixed as 'doing treatment'. It is possible that this content was too abstractly and/or comprehensively formulated for an effect to occur.

**Differences in goal regulation between adolescents with CF or diabetes and their healthy peers**

Additional research should further investigate possible differences in goal setting and striving between adolescents with and without chronic illness. The current lack of findings does not suggest a generalizable absence of differences as it depends on statistical power. Therefore, future research in larger samples is needed.

This research should also try to obtain higher response rates, in order to obtain more representative samples of adolescents with CF or diabetes. Given the rather low response rates in the current set of studies, it is possible that the participating adolescents possess certain characteristics that do not generalize to non-participating adolescents and young adults (e.g., relatively good health, less disability, less hindrance, etc.).

Also, the comparison between chronically ill and healthy adolescents should also be made for other chronic conditions such as epilepsy, asthma, arthritis, etc.

Additionally, it would also be interesting to focus on how adolescents and young adults with CF or diabetes manage to incorporate or reconcile their health and non-health goals. Understanding which strategies these adolescents and young adults use to manage this may be essential in the adaptation to their illness. These strategies may include the use of compatible strategies to attain different goals (e.g., 'join a running team' may be effective for both 'obtain a better lung functioning' and 'meet new people'), the identification of possible instrumental relations between goals (e.g., 'take responsibility for treatment' may be a step towards 'become independent'), the optimization of the use of limited resources (e.g., time, energy; e.g., 'doing aerosol treatment while studying'), the adoption of more realistic goals, and/or the adjustment of goals to situational constraints (e.g., obtain a college instead of a university degree) (Brandstätter & Renner, 1990; Riediger & Freund, 2004; Schwartz & Drotar, 2009).

Finally, longitudinal studies investigating how adolescents with chronic illness, from the time of diagnosis on, set, pursuit, adjust, attain, and balance their health and non-health goals may be interesting to fully understand their adjustment to illness, and to promote disease management, and future physical health.

**Goals in adolescents with chronic illness**

Despite the recently growing attention for the investigation of goals in adolescents with chronic illness, existing research concerning this topic is still scarce. Goals are especially important for adolescents with chronic illness because of the need to achieve important life tasks in the transition to adulthood, a growing autonomy in the

regulation of their goals, their cognitive capability to evaluate the impact of illness on goal regulation, their desire to be 'normal', their tendency to be non-adherent to treatment, and the importance of good disease management (Badlan, 2006; Little, 1983; Massey, et al. 2008; Schwartz & Drotar, 2006, 2009). Studies focusing on goals have already been conducted in adolescents with diabetes, asthma, cerebral palsy, phenylketonuria, cancer, and CF (Helgeson & Takeda, 2009; for a review see Schwartz & Drotar, 2006; Schwartz & Drotar, 2009; Schwartz & Parisi, in press; Seiffge-Krenke, 1998). These studies primarily focused on treatment and adherence goals, but also compared goal setting and striving between adolescents with and without chronic illness. These studies should be replicated and extend by also examining the interplay between health and non-health goals. Especially, attention should be paid to the experience of intergoal interference and the interference of goals by health, and their relationship with, not only well-being, but also treatment adherence.

Another direction for future research is to study how adolescents with chronic illness select their goals in an environment with multiple goals en how they shield the pursuit of these goals from other competing goals. Interesting may be to investigate which determinants (e.g., desirability, perceived difficulty or feasibility, self-efficacy, etc.) play a role in the selection of goals of adolescents with chronic illness (Gollwitzer & Oettingen, 2012). Knowing how these adolescents select their goals does not imply that they will effectively attain these goals. Attaining a goal also depends and how one pursues these goals and on how one shields or protects this goal pursuit from getting derailed (Gollwitzer & Oettingen, 2012). Situations that may cause threats to goal pursuit are for example attentional limitations (e.g., distractions), unexpected obstacles and barriers, certain inner states (e.g., craving, mood, anxiety, etc.), and other competing goals (Achtziger, Gollwitzer, & Sheeran, 2008). These situations may especially arise in adolescents with chronic illness (Michaud et al., 2004). Therefore, examining goal shielding in the context of chronic illness may be of high relevance. Goal shielding refers to pushing down attractive alternative goals that may occur as contexts change (Shah & Kruglanski, 2007). Recent research suggests that implementation intentions (i.e., 'IF… THEN…' plans) may prove useful in to keep goal pursuit on track (see Gollwitzer & Oettingen, 2012). Future research may investigate whether these implementation intentions are beneficial in the context of chronic illness.

Furthermore, next to investigating goal setting and striving from an intrapersonal perspective, research may also examine goals in the context of chronic illness from an

interpersonal perspective. As adolescence is a period where autonomy and independence in goal regulation should be developed and parental involvement should decrease, investigating the influence of significant others on the goals of these adolescents is of great importance. This research should not only focus on the (health and non-health) goals that parents have for their adolescent but also on the goals that physicians have for this adolescent. Recent research indeed stresses the need to take into account the adolescent-parent-physician triad concerning decision making and goal setting in the context of chronic illness (Lipstein, Brinkman, & Britto, 2012). This research could focus on the level of conflict between the goals that these adolescents have and the goals their parent(s)/physician has/have for them. Interesting would be to investigate whether the level of this conflict relates to outcomes such as treatment adherence and well-being. This research would extend recent research concerning family conflict and adherence in adolescents with diabetes (e.g., Herzer, Vesco, Ingerski, Dolan, & Hood, 2011), and recent research concerning the accuracy of parents to predict the goals of their adolescent with diabetes (e.g., Butler et al., 2012). Furthermore, building further on recent research concerning autonomy and independence in the pursuit of health goals (Butner et al., 2009; Devine, Wasserman, Gershenson, Holmbeck, & Essner, 2011; Miller & Harris, 2012), investigating adolescents' and their parents' perceptions of autonomy and independence concerning adolescents' health and non-health goals, and the willingness of parents to allow this autonomy and independence would be useful.

## REFERENCES

Achtziger, A., Gollwitzer, P.M., & Sheeran, P. (2008). Implementation intentions and shielding goal striving from unwanted thoughts and feelings. *Personality and Social Psychology Bulletin, 34,* 381-393. doi: 10.1177/0146167207311201

Bach, P., & Hayes, S.C. (2002). The use of acceptance and commitment therapy to prevent the rehospitalization of psychotic patients: a randomized controlled trial. *Journal of Consulting and Clinical Psychology, 70(5),* 1129-1139. doi: 10.1037//0022-006X.70.5.1129

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Brandtstädter, J. & Renner, G. (1990). Tenacious goal pursuit and flexible goal adjustment: explication and age-related analysis of assimilative and accommodative strategies of coping. *Psychology and Aging, 5,* 58-67.

Butler, J.M., Fortenberry, K.T., Berg, C.A., McCabe, J., Blakemore, T., & Wiebe, D.J. (2011). Early adolescents' goals and mothers' accuracy in predicting adolescent goals in the context of Type 1 diabetes. *Childrens' Health Care, 40,* 155-169. doi: 10.1080/02739615.2011.564940

Butner, J., Berg, C.A., Osborn, P., Butler, J.M., Godri, C., Fortenberry, K.T.,…Wiebe, D.J. (2009). *Developmental Psychology, 45,* 835-849. doi: 10.1037/a0015363

Ciarrochi, J. Bilich, L., & Godsel, C. (2010). Psychological flexibility as a mechanism of change in Acceptance and Commitment Therapy. In R. Baer (Ed.), *Assessing Mindfulness and Acceptance: Illuminating the Processes of Change* (pp. 51-76). New Harbinger Publications, Inc.: Oakland, CA.

Cole, D.A. & Maxwell, S.E. (2003). Testing mediation models with longitudinal data: questions and tips in the use of structural equation modeling. *Journal of Abnormal Psychology, 112,* 558-577. doi: 10.1037/0021-843X.112.4.558

Damme, S. Van, Crombez, G., Houdenhove, B. Van, Mariman, A., & Michielsen, W. (2006). Well-being in patients with chronic fatigue syndrome: The role of acceptance. *Journal of Psychosomatic Research, 61,* 595-599. doi:10.1016/j.jpsychores.2006.04.015

Devine, K.A., Wasserman, R.M., Gershenson, L.S., Holmbeck, G.N., & Essner, B.S. (2011). Mother-adolescent agreement regarding decision-making autonomy: a longitudinal comparison of families of adolescents with and without spina bifida. *Journal of Pediatric Psychology, 36,* 277-288. doi: 10.1093/jpepsy/jsq093

Ernst, M.M., Johnson, M.C., & Stark, L.J. (2010). Developmental and psychosocial issues in cystic fibrosis. *Child and Adolescent Psychiatric Clinics of North America, 19,* 263-283. doi: 10.1016/j.chc.2010.01.004

Evers, A.W.M., Kraaimaat, F.W., van Langenveld, W., Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Feinstein, A.B., Forman, E.M., Masuda, A., Cohen, L.L., Herbert, J.D., Moorthy, L.N., & Goldsmith, D.P. (2011). Pain intensity, psychological inflexibility, and acceptance of pain as predictor of functioning in adolescents with juvenile

idiopathic arthritis: a preliminary investigation. *Journal of Clinical Psychology in Medical Settings, 18,* 291-298. doi: 10.1007/s10880-011-9243-6

Gauthier, L.R., Roding, G., Zimmerman, C., Warr, D., Moore, M., Shepherd, F., & Gagliese, L. (2009). Acceptance of pain: A study in patients with advanced cancer. *Pain, 143,* 147-154. doi:10.1016/j.pain.2009.02.009

Gjengedal, E., Rustøen, T., Wahl, A.K., & Hanestadt, B.R. (2003). Growing up and living with cystic fibrosis. Everyday life and encounters with the health care and social services – A qualitative study. *Advances in Nursing Science, 26 (2),* 149-159.

Glasscoe, C.A. & Quittner, A.L. Psychological interventions for people with cystic fibrosis and their families. *Cochrane Database of Systematic Reviews* 2008, Issue 3. Art. No.: CD003148. doi: 10.1002/14651858.CD003148.pub2.

Gollwitzer, P.M. & Oettinger, G. (2012). Goal Pursuit. In R.M. Ryan (Ed.), *The Oxford Handbook of Human Motivation* (pp. 208-231). Oxford University Press Inc.: New York, New York.

Hayes, S.C. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapies. *Behavior Therapy, 35,* 639-665. doi: 10.1016/S00005-7894(04)80013-3

Hayes, S.C., Bissett, R.T., Korn, Z., Zettle, R.D., Rosenfarb, I.S., Cooper, L.D., & Grundt, A.M. (1999). The impact of acceptance versus control rationales on pain tolerance. *The Psychological Record, 49,* 33-47.

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: model, processes and outcomes. *Behaviour Research and Therapy, 44,* 1-25. doi:10.1016/j.brat.2005.06.006

Heffner, M., Sperry, J., Eifert, G.H., & Detweiler, M. (2002). Acceptance and Commitment Therapy in the treatment of an adolescent with anorexia nervosa. A case example. *Cognitive and Behavioral Practice, 9,* 232-236. doi:10.1016/S1077-7229(02)80053-0

Helgeson, V. S., & Takeda, A. (2009). Brief report: Nature and implications of personal projects among adolescents with and without diabetes. *Journal of Pediatric Psychology, 34,* 1019–1024. doi:10.1093/jpepsy/jsp024

Herzer, M., Vesco, A., Ingerski, L.M., Dolan, L.M., & Hood, K.K. (2011). Explaining family conflict-glycemic control link through psychological variables in

adolescents with type 1 diabetes. *Journal of Behavioral Medicine, 34,* 268-274. doi: 10.1007/s10865-010-9307-3

Lipstein, E.A., Brinkman, W.B., & Britto, M.T. (2012). What is known about parents' treatment decisions? A narrative review of pediatric decision making. *Medical Decision Making, 32,* 246-258. doi: 10.1177/0272989X11421528

Little, B. R. (1983). Personal projects: A rationale and method for investigation. *Environment and Behavior, 1*5, 273–309.

Maes, S. & Karoly, P. (2005). Self-regulation assessment and intervention in physical health and illness: a review. *Applied Psychology: An International Review, 54(2),* 267-299.

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,* 421-460. doi: 10.1016/j.dr.2008.03.002

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2009a). Self-generated goals and goal process appraisals: Relationships with sociodemographic factors and well-being. *Journal of adolescence, 32,* 501-518. doi: 10.1016/j.adolescence.2008.07.003

Massey, E.K., Garnefski, N., Gebhardt, W.A., & van der Leeden, R. (2009b). Daily frustration, cognitive coping and coping efficacy in adolescent headache: a daily diary study. *Headache, 49,* 1198-1205. doi: 10.1111/j.1526-4610.2009.01492.x

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: Acceptance and Commitment Therapy for pediatric sickle cell disease. *Journal of Pediatric Psychology, 36,* 398-408. doi:10.1093/jpepsy/jsq118

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents and young adults with chronic pain: validation of an adapted assessment instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi:10.1016/j.ejpain.2009.05.002

McCracken, L.M. & Vowles, K.E. (2008). A prospective analysis of acceptance of pain and values-based action in patients with chronic pain. *Health Psychology, 27(2),* 215-220. doi: 10.1037/0278-6133.27.2.215

McCracken, L.M. & Zhao-O'Brien, J. (2010). General psychological acceptance and chronic pain: There is more to accept than the pain itself. *European Journal of Pain, 14(2),* 170-175. doi:10.1016/j.ejpain.2009.03.004

Michaud, P.A., Suris, J.C., & Viner, R. (2004). The adolescent with a pediatric condition. Part II: healthcare provision. *Archives of Disease in Childhood, 89,* 943-949. doi: 10.1136/adc.2003.045377

Miller, V.A. & Harris, D. (2012). Measuring children's decision-making involvement regarding illness management. *Journal of Pediatric Psychology, 37,* 292-306. doi: 10.1093/jpepsy/jsr097

Nurmi, J.E. (1987). Age, sex, social class, and quality of family interactions as determinants of adolescents' future orientation: A developmental task interpretation. *Adolescence, 22,* 977–991.

Nurmi, J.E. (1991). How do adolescents see their future? A review of the development of future orientation and planning. *Developmental Review, 11,* 1–59.

Nurmi, J.E. (1993). Adolescent development in an age-graded context: The role of personal beliefs, goals, and strategies in the tackling of developmental tasks and standards. *International Journal of Behavioral Development, 16,* 169–189.

Pakenham, K.I. (2006). Investigation of the coping antecedents to positive outcomes and distress in multiple sclerosis (MS). *Psychology and Health, 21(5),* 633-649. doi: 10.1080/14768320500422618

Pinquart, M., & Shen, Y. (2011). Behavior problems in children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 36,* 1003-1016. doi: 10.1093/jpepsy/jsr042

Pinquart, M., & Teubert, D. (2012). Academic, physical, and social functioning of children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 37,* 376-389. doi: 10.1093/jpepsy/jsn06

Riediger, M. (2007). Interference and facilitation among personal goals: Age-group differences and associations with well-being and behavior. In B.R. Little, K. Salmela-Aro, J.-E. Nurmi, & S.D. Philipps (Eds.), *Personal project pursuit: Goals action, and human flourishing* (pp. 119-143). Erlbaum.

Riediger, M. & Freund, A.M. (2004). Interference and facilitation among personal goals: differential associations with subjective well-being and persistent goal pursuit. *Personality and Social Psychology Bulletin, 30,* 1511-1523. doi: 10.1177/0146167204271184

Risdon, A., Eccleston, C., Crombez, G., & McCracken, L. (2003). How can we learn to live with the pain? A Q-methodological analysis of the diverse understandings of

acceptance of chronic pain. *Social Science and Medicine, 56,* 375-386. doi: 10.1016/S0277-9536(02)00043-6

Schwartz, L.A. & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 393-405. doi: 10.1007/s10880-006-9041-8

Schwartz, L.A. & Drotar, D. (2009). Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 34,* 954-965. doi: 10.1093/jpepsy/jsn144

Schwartz, L.A. & Parisi, M.L. (in press). Self-identified goals of adolescents with cancer and healthy peers: content, appraisals, and correlates. *Journal of Pediatric Psychology.*

Seiffge-Krenke, I. (1998). Chronic disease and perceived developmental progression in adolescence. *Developmental Psychology, 34,* 1073–1084. doi:10.1037/0012-1649.34.5.1073

Seiffge-Krenke, I. (2001). Diabetic adolescents and their families: stress, coping, and adaptation. Cambridge, UK: Cambridge University Press.

Shah, J.Y., & Kruglanski, A.W. (2007). Structural dynamics. The challenge of change in goal systems. In J.Y. Shah & Gardner W.L. (Eds.), *Handbook of Motivation Science* (pp. 217-229). The Guilford Press: New York, New York.

Westin, V., Hayes, S.C., & Andersson, G. (2008). Is it the sound or your relationship to it? The role of acceptance in predicting tinnitus impact. *Behaviour Research and Therapy, 46,* 1259-1265. doi:10.1016/j.brat.2008.08.008

Wicksell, R.K., Dahl, J., Magnusson, B., & Olsson, G. (2005). Using acceptance and commitment therapy in the rehabilitation of an adolescent female with chronic pain: a case example. *Cognitive and Behavioral Practice, 12,* 415-423. doi: 10.1016/S1077-7229(05)80069-0

Wicksell, R.K., Melin, L., Lekander, M., & Olsson, G.L. (2009). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – a randomized controlled trial. *Pain, 141,* 248-257. doi: 10.1016/j.pain.2008.11.006

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi:10.1016/j.pain.2011.09.003

Yeo, M. & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330,* 721-723. doi: 10.1136/bmj.330.7493.721

# NEDERLANDSTALIGE SAMENVATTING

## INLEIDING

Voor adolescenten met een chronische ziekte kan de continue gezondheidsdreiging in combinatie met een veeleisende behandeling een sterk negatieve invloed hebben op hun welzijn. Voorgaand onderzoek toonde inderdaad aan dat opgroeien met een chronische ziekte een risico vormt voor verschillende moeilijkheden zoals depressieve symptomen, gedragsproblemen, en achteruitgang in hun academisch, fysiek, en sociaal functioneren (see Pinquart & Shen, 2011; Pinquart & Teubert, 2012; Schwartz & Drotar, 2006, 2009; Yeo & Sawyer, 2005). Daarom is het belangrijk om te begrijpen hoe deze adolescenten hun welzijn op peil kunnen houden ondanks de last van het ziek zijn .

Binnen dit proefschrift werd er gefocust op 'aanvaarding van ziekte' als mogelijke beschermende factor voor het welzijn van adolescenten met een chronsiche ziekte (Evers et al., 2001). Immers, voorgaand onderzoek heeft reeds aangetoond dat aaanvaarding inderdaad een beschermende rol kan spelen in het welzijn van adolescenten met verschillende aandoeningen zoals chronische pijn (e.g., McCracken, Gauntlett-Gilbert, & Eccleston, 2010; Wicksell, Olssen, & Hayes, 2011), juveniele arthritis (Feinstein et al., 2011), en sikkelcel ziekte (Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011). Dit onderzoek toonde aan dat meer aanvaarding gerelateerd is aan positieve uitkomsten zoals minder angst, minder depressie, minder hinder, en een beter emotioneel, sociaal, en fysiek welzijn.

Naast het onderzoeken van de rol van aanvaarding in het welzijn van deze adolescenten, is ook het begrijpen van het proces waarlangs aanvaarding welzijn beïvloedt van belang. Dit proefschrift richtte zich op twee spedicieke doelregulatie processen (i.e., doel interferentie en doel facilitatie) als mogelijke mediatoren. Deze doelregulatie processen zijn uitermate van belang voor adolescenten met chronische ziekte. Voor deze adolescenten is het immers belangrijk om essentiële levensdoelen te bereiken zodat overgegaan kan worden naar de volwassenheid. Bovendien beginnen zij hun doelen meer autonoom te reguleren, zijn ze cognitief in staat om de gevolgen van hun ziekte op hun doelregulatie in te zien, hebben ze een sterke wens hebben om 'normaal' te zijn, zijn ze geneigd om zich niet aan hun behandeling te houden, en zijn ze genoodzaakt

om behandelingsdoelen na te streven die kunnen interfereren met het nastreven van andere levensdoelen (e.g., onafhankelijk worden) (Badlan, 2006; Little, 1983; Massey, Gebhardt, & Garnefski, 2008; Nurmi, 1987, 1991,1993; Schwartz & Drotar, 2006, 2009).

### DOELSTELLING

Dit proefschrift heeft als doel het onderzoeken van (1) de rol van aanvaarding van ziekte in het welzijn van adolescenten met chronische ziekte, (2) de rol van aanvaarding van ziekte in het ervaren van doel interferentie en facilitatie van deze adolescenten, (3) de mediërende rol van doel interferentie en facilitatie in de relatie tussen aanvaarding en welzijn, en (4) de verschillen in doelregulatie tussen adolescenten met een chronische ziekte en gezonde leeftijdsgenoten. Deze doelstellingen werden onderzocht aan de hand van zes studies, gebruik makend van verschillende onderzoeksmethoden, bij adolescenten met muscoviscidose en diabetes, en hun gezonde leeftijdsgenoten. Welzijn werd geoperationaliseerd in termen van positief affect, negatief affect, en levenssatisfactie (Diener, 1984; Diener, Suh, Lucas, & Smith, 1999). Positief affect verwijst naar stemmingen en emoties zoals blijheid, geluk, en tevredenheid, terwijl negatief affect emotionele responsen zoals angst, depressie, en stress omvat (Diener et al., 1999). Levenssatisfactie, als derde component, houdt een globale evaluatie van iemands leven over verschillende domeinen in (Diener, 1984; Diener et al., 1999).

### RESULTATEN

**De rol van aanvaarding van ziekte in het welzijn van adolescenten met een chronische ziekte**

**Hoofstuk 1** rapporteert de bevindingen van een cross-sectionele vragenlijststudie die de relatie tussen aanvaarding en een eerste component van welzijn (i.e., negatief affect) bestudeerde. Aan deze studie namen 34 adolescenten met mucoviscidose tussen 12 en 22 jaar deel. Alle adolescenten vulden een batterij vragenlijsten in die aanvaarding, angst, depresieve symptomen, en functionele hinder meten. In lijn met de verwachtingen, suggereerden de resultaten van deze studie dat meer aanvaarding van ziekte gerelateerd was aan minder angst, depressieve symptomen, en functionele hinder bovenop de effecten van ziekte-ernst. **Hoofdstuk 2** had als doel om deze bevindingen te repliceren, maar breidde ze ook uit door gebruik te maken van een prospectief design en door ook te focussen op de relatie tussen aanvaarding en een derde component van welzijn: levenssatisfactie. Veertig adolescenten met mucoviscidose (14-22 jaar) namen aan deze

prospectieve vragenlijstenstudie deel. Aanvaarding, angst, depressieve symptomen, en fysieke, emotionele, rol, en social functioneren werden gemeten aan de hand van zelf-rapportage vragenlijsten. Deze vragenlijsten werden ingevuld op een eerste meetmoment (Tijdstip 1) en zes maanden later (Tijdstip 2). Opnieuw werd er gevonden dat meer aanvaarding van ziekte gerelateerd was aan minder depressieve symptomen (op Tijdstip 1 en 2) maar, tegengesteld aan de verwachtingen, niet aan angst. Verder suggereerden de bevindingen dat meer aanvaarding samenhing met een beter emotioneel, rol, en sociaal fucntioneren op Tijdstip 1, maar niet op Tijdstip 2. Aanvaarding was echter niet gerelateerd aan fysiek functioneren. In **hoofdstuk 3** onderzochten we opnieuw de relatie tussen aanvaarding en negatief affect. Verschillend van hoofdstuk 1 en 2, werd negatief affect gemeten tijdens een reële situatie. Zesendertig adolescenten (12-22 jaar) vulden een vragenlijst die aanvaarding meet in en rapporteerden in welke mate ze pijn en stress verwachten en ervaren tijdens een medische procedure die ze moesten ondergaan in het ziekenhuis (i.e., longfunctiemeting). Ook fysiologische maten van stress werden geregistreerd (i.e., hartslag en hartslagvariabiliteit). De bevindingen van deze studie toonden aan dat meer aanvaarding van ziekte gerelateerd was aan minder verwachte pijn en pijngerelateerde gedachten, maar niet aan de mate van ervaren stress voor en tijdens de longfucntiemeting, noch aan de mate van ervaren pijn.

**Verschillen in doelregulatie tussen adolescenten met een chronische ziekte en gezonde leeftijdsgenoten**

In **hoofdstuk 4** werd doelregulatie geïntroduceerd. Doelen en doelregulatieprocessen werden vergeleken tussen adolescenten met ($N = 60$; 14-22 jaar) en zonder chronische ziekte ($N = 60$; 14-22 jaar), gemeten aan de hand van vragenlijsten. Er werd gevonden dat adolescenten met chronische ziekte hun gezondheidsdoelen als meer belangrijk en moeilijker te bereiken beoordeelden in vergelijking met hun gezonde leeftijdsgenoten. Tegengesteld aan onze verwachtingen beoordeelden adolescenten met chronische ziekte zichzelf als meer succesvol in het bereiken van deze gezondheidsdoelen. Verder werden er geen verschillen teruggevonden tussen adolescenten met en zonder ziekte wat betreft het belang, de moeilijkheid en succes van hun niet-gezondheidsdoelen, behalve voor de moeilijkheid in het bereiken van relatie doelen, wat als minder moeilijk werd beoordeeld door de adolescenten met chronische ziekte. Uiteindelijk werden ook geen verschillen gevonden in de mate van ervaren interferentie in niet-gezondheidsdoelen door gezondheidsdoelen teruggevonden tussen beide groepen.

**De rol van aanvaarding van ziekte in het ervaren van doel interferentie en facilitatie van deze adolescenten en de mediërende rol van doel interferentie en facilitatie in de relatie tussen aanvaarding en welzijn**

**Hoofdstuk 5** ging verder met het onderzoeken van doelregulatie processen door na te gaan of dagelijks ervaren doelinterferentie en –facilitatie van levensdoelen door behandelingsdoelen bij adolescenten met mucoviscidose en diabetes de relatie tussen aanvaarding van ziekte en welzijn medieert. Deze studie werd uitgevoerd bij 38 adolescenten met mucoviscidose en diabetes (14-22 jaar) en maakte gebruik van een dagelijkse proces analyse. Naast het bevragen van aanvaarding van ziekte, negatieve levensgebeurtenissen, en doelgerelateerde eigen-effectiviteit door middel van vragenlijsten, werden dagelijkse stemming en dagelijks ervaren doelinterferentie en –facilitatie van levensdoelen door behandelingsdoelen bevraagd in een online dagboek. Dit dagboek diende ingevuld te worden gedurende drie opeenvolgende weken. De resultaten van deze studie toonden aan dat aanvaarding van ziekte positief gerelateerd was aan dagelijks welzijn, maar niet aan dagelijks ervaren doelinterferentie en –facilitatie. Verder werd gevonden dat dagelijks ervaren doelinterferentie en –facilitatie niet gerelateerd waren aan welzijn gemeten op dezelfde en de volgende dag. In **hoofdstuk 6** beoogden we om de bevindingen van hoofdstuk 5 te repliceren gebruik makend van een andere methodologie. Ditmaal werd gebruik gemaakt van een longitudinaal, in plaats van dagelijks proces, design. Dertig participanten met mucoviscidose en diabetes vulden een set vragenlijsten in die aanvaarding van ziekte, negatieve levensgebeurtenissen, doelgerelateerde eigen-effectiviteit, welzijn, en doelinterferentie en –facilitatie van levensdoelen door behandelingsdoelen meten. Deze vragenlijsten werden op drie opeenvolgende momenten ingevuld (baseline, twee, en vier maanden later). Opnieuw werd er gevonden dat aanvaarding van ziekte (op Tijdstip 1) positief gerelateerd was aan een aantal dimensies van welzijn (op Tijdstip 3; i.e., anst en depressieve symptomen, maar niet levenskwaliteit). Verder bleek dat aanvaarding van ziekte (op Tijdstip 1) niet gerelateerd was aan doelinterferentie en –facilitatie (op Tijdstip 2). Ook waren doelinterferentie en –facilitatie (op Tijdstip 2) niet gerelateerd aan de dimensies van welzijn (op Tijdstip 3). Hoofdstuk 6 bevestigde dus de resultaten gevonden in hoofdstuk 5.

## DISCUSSIE

Op basis van de bevindingen uit dit proefschrift kan geconcludeerd worden dat aanvaarding van ziekte gerelateerd lijkt te zijn aan aanpassing aan ziekte in het algemeen (zie hoofdstuk 1, 2, 5, en 6), maar ook aan specifieke ziektegerelateerde ervaringen zoals verwachte pijn gedurende medische procedures (i.e., longfunctiemeting) en pijngerelateerde gedachten (zie hoofdstuk 3). Verder werd gevonden dat aanvaarding van ziekte niet gerelateerd was aan doel interferentie en –facilitatie, en de relatie tussen aanvaarding en welzijn dus niet gemedieerd werd door deze doelregulatie processen (zie hoofdstuk 5 en 6). Uiteindelijk bleek ook dat het stellen en nastreven van doelen grotendeels gelijk loopt bij adolescenten met en zonder een chronische ziekte, uitgezonderd wat betreft het ervan belang en de moeilijkheid in het bereiken van hun gezondheidsdoelen.

## KLINISCHE IMPLICATIES

De resultaten van dit proefschrift suggereren dat adolescenten met een chronische ziekte voordeel kunnen halen uit interventies die aanvaarding van ziekte promoten. Deze interventies kunnen hun welzijn ondersteunen en/of verhogen wanneer het aangetast is. Interventies die focussen op aanvaarding, zoals Acceptance and Commitment Therapy (ACT; Hayes, 2004) zijn reeds ontwikkeld voor kinderen en adolescenten met sikkelcel ziekte en chronische pijn en lijken te leiden tot minder symptomen, een beter functioneren, en een betere levenskwaliteit (e.g., Masuda et al., 2011; Wicksell, Melin, Lekander, & Olsson, 2009). Verder onderzoek is nodig om te achterhalen of ACT ook zinvol kan zijn voor adolescenten met mucoviscidose en diabetes. Indien dit het geval is, kunnen bestaande interventies aangepast worden voor en uitgetest worden bij deze specifieke populatie.

## REFERENTIES

Badlan, K. (2006). Young people living with cystic fibrosis: an insight into their subjective experience. *Health and Social Care in the Community, 14(3),* 264-270. doi: 10.1111/j.1365-2524.2006.00619.x

Diener, E. (1984). Subjective well-being. *Psychological Bulletin, 95(3),* 542-575.

Diener, E., Suh, E.M., Lucas, R.E., & Smith, H.L. (1999). Subjective well-being: Three decades of progress. *Psychological Bulletin, 125,* 276-302.

Evers, A.W.M., Kraaimaat, F.W., van Langenveld, W., Jongen, P.J.H., Jacobs, J.W.G., & Bijlsma, J.W.J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69,* 1026-1036. doi: 10.1037//0022-006X.69.6.1026

Feinstein, A.B., Forman, E.M., Masuda, A., Cohen, L.L., Herbert, J.D., Moorthy, L.N., & Goldsmith, D.P. (2011). Pain intensity, psychological inflexibility, and acceptance of pain as predictor of functioning in adolescents with juvenile idiopathic arthritis: a preliminary investigation. *Journal of Clinical Psychology in Medical Settings, 18,* 291-298. doi: 10.1007/s10880-011-9243-6

Hayes, S.C. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapies. *Behavior Therapy, 35,* 639-665. doi: 10.1016/S00005-7894(04)80013-3

Little, B. R. (1983). Personal projects: A rationale and method for investigation. *Environment and Behavior, 1*5, 273–309.

Massey, E.K., Gebhardt, W.A., & Garnefski, N. (2008). Adolescent goal content and pursuit: a review of the literature from the past 16 years. *Developmental Review, 28,* 421-460. doi: 10.1016/j.dr.2008.03.002

Masuda, A., Cohen, L.L., Wicksell, R.K., Kemani, M.K., & Johnson, A. (2011). A case study: Acceptance and Commitment Therapy for pediatric sickle cell disease. *Journal of Pediatric Psychology, 36,* 398-408. doi:10.1093/jpepsy/jsq118

McCracken, L.M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents and young adults with chronic pain: validation of an adapted assessment instrument and preliminary correlation analyses. *European Journal of Pain, 14,* 316-320. doi:10.1016/j.ejpain.2009.05.002

Nurmi, J.E. (1987). Age, sex, social class, and quality of family interactions as determinants of adolescents' future orientation: A developmental task interpretation. *Adolescence, 22,* 977–991.

Nurmi, J.E. (1991). How do adolescents see their future? A review of the development of future orientation and planning. *Developmental Review, 11,* 1–59.

Nurmi, J.E. (1993). Adolescent development in an age-graded context: The role of personal beliefs, goals, and strategies in the tackling of developmental tasks and standards. *International Journal of Behavioral Development, 16,* 169–189.

Pinquart, M., & Shen, Y. (2011). Behavior problems in children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 36,* 1003-1016. doi: 10.1093/jpepsy/jsr042

Pinquart, M., & Teubert, D. (2012). Academic, physical, and social functioning of children and adolescents with chronic physical illness: a meta-analysis. *Journal of Pediatric Psychology, 37,* 376-389. doi: 10.1093/jpepsy/jsn06

Schwartz, L.A. & Drotar, D. (2006). Defining the nature and impact of goals in children and adolescents with a chronic health condition: review of research and a theoretical framework. *Journal of Clinical Psychology in Medical Settings, 13,* 393-405. doi: 10.1007/s10880-006-9041-8

Schwartz, L.A. & Drotar, D. (2009). Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *Journal of Pediatric Psychology, 34,* 954-965. doi: 10.1093/jpepsy/jsn144

Wicksell, R.K., Melin, L., Lekander, M., & Olsson, G.L. (2009). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – a randomized controlled trial. *Pain, 141,* 248-257. doi: 10.1016/j.pain.2008.11.006

Wicksell, R.K., Olsson, G.L., & Hayes, S.C. (2011). Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain, 152,* 2792-2801. doi:10.1016/j.pain.2011.09.003

Yeo, M. & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330,* 721-723. doi: 10.1136/bmj.330.7493.721

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Annabelle