# Fears, Stress and Burnout in Parents of Children with Chronic Conditions

Treatment with Cognitive Behavioural Therapy and Mindfulness

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## ABSTRACT

The aim of the present research was threefold: to investigate the fears of parents of children with chronic conditions who suffer from fears, stress and burnout; to evaluate the effectiveness of their treatment with either mindfulness-based therapy or cognitive behavioural therapy (CBT); and to assess treatment outcome in terms of health-related quality of life (HRQoL). Research on parents of children with chronic conditions has shown that this parent group frequently suffers from psychological problems. Long-term stress can lead to some form of chronic stress reaction. In study one, parents of children with brain tumours were asked to rate the extent to which they experienced a set of specific fears related to their child's brain tumour and its treatment. Fears of future cancer recurrence and of late effects of treatment were most prominent among parents of CNS tumour patients. Study two investigated the effectiveness of two group-based interventions on stress and burnout among parents of children with chronic conditions. After a waiting list control period, parents were offered either a CBT or a mindfulness programme. After eight group therapy sessions, both interventions significantly decreased stress and burnout. Study three focused on the HRQoL and life satisfaction of the parents in study two. The results indicate improvements for participants in both treatment groups regarding certain areas of HRQoL and life satisfaction. To conclude, many parents of children with chronic conditions suffer from stress-related mental illness and need targeted interventions for their own problems. The present research concludes that fears concerning future cancer recurrence and concerning late effects of treatment are most prominent among parents of children with cancer. Another conclusion is that CBT and mindfulness decrease stress and burnout and may have a positive effect on areas of HRQoL and life satisfaction in parents of children with chronic conditions.

Key words: Parents of children with chronic conditions, Mindfulness, CBT, stress, burnout, HRQoL, life satisfaction

## SUMMARY IN SWEDISH – SAMMANFATTNING

Det huvudsakliga syftet med denna avhandling var att kartlägga rädslor och undersöka om kognitiv beteendeterapi (KBT) och Mindfulness kan reducera stress och utmattningssymtom hos föräldrar till barn med kronisk sjukdom samt undersöka om KBT och Mindfulness förbättrar föräldrarnas livskvalitet. Tidigare forskning gällande föräldrar till barn med kronisk sjukdom visar att denna föräldragrupp lider av både rädslor, stress och utmattningsrelaterade besvär samt försämrad livskvalitet. Långvarig stress kan efterhand övergå i någon form av kronisk stressreaktion eller utmattningstillstånd. Studie I undersöker upplevda rädslor hos föräldrar till barn som drabbats av hjärntumör ($n = 82$). Resultatet jämförs sedan med föräldrar till barn med akut lymfatisk leukemi (ALL; $n = 208$) och analyseras i relation till behandlingssituation och tid sedan barnet fick sin diagnos. Resultaten visade att föräldrar, oavsett barnets diagnos, lider av rädslor i hög grad. Rädsla för återfall och sena komplikationer var de mest framträdande rädslorna hos båda grupperna, men föräldrar till barn med hjärntumör skattade dessa rädslor högre. Dessutom uppger ungefär en fjärdedel av dessa föräldrar att de är rädda för att det ska "gå helt utför" för barnet. Det återkommande behovet av kontroller och möjliga tecken på återfall gör att föräldrarna upplever en stor osäkerhet om det slutgiltiga utfallet av sjukdomen, trots att den aktiva behandlingen sedan länge avslutats. Studie II undersökte om gruppbehandling med KBT respektive Mindfulness kan vara effektiva metoder för att lindra stress och utmattningssymtom hos föräldrar till barn med kronisk sjukdom som lider av stress och utmattningsproblematik. Efter att först ha stått på väntelista i 6 månader lottades föräldrarna ($n = 21$) till gruppbehandling med KBT ($n = 10$) eller Mindfulness ($n = 9$). Båda gruppbehandlingarna minskade stress och utmattningssymtom signifikant. Effektstorleken var stor för båda grupperna (KBT, $g = 1,28–1,64$; Mindfulness, $g = 1,25–2,20$). Studie III var en förstudie som fokuserade på hälsorelaterad livskvalitet (HRQoL) och nöjdhet med livet. Efter åtta veckors behandling hade såväl föräldrarnas övergripande Psykiska hälsa (MCS) som värdena på de mentala delskalorna vitalitet, social funktion, emotionell rollfunktion, och psykiskt välbefinnande förbättrats signifikant för båda grupperna (KBT och Mindfulness). Dessutom förbättrades värdena på de fysiska delskalorna fysisk rollfunktion, kroppslig smärta och allmän hälsoupplevelse hos Mindfulnessgruppen. Klinisk signifikans testades genom att jämföra studiepopulationens medelvärden med medelvärden från en redan framtagen normgrupp. Det visade sig att MCS-värdet var betydligt lägre före behandling i

studiepopulationen men inga signifikanta skillnader fanns kvar efter genomförd behandling. För den övergripande Fysiska hälsan (PCS) noterades en signifikant högre poäng efter behandling jämfört med normgruppen. Resultaten indikerar även förbättrad nöjdhet med livet, särskilt med avseende på egentid och relationen med barnet och partnern. Sammanfattningsvis lider många föräldrar till barn med kronisk sjukdom av stress och utmattningsproblematik och är i behov av riktade interventioner för sina egna problem. Denna avhandling visar att rädslan för återfall och sena komplikationer är vanliga hos föräldrar till barn med cancer – särskilt hos föräldrar till barn som behandlats för hjärntumör. Avhandlingen visar också att såväl KBT för stress och utmattning som Mindfulness minskar stress och utmattningssymtom och kan ha en positiv effekt med avseende på HRQoL och nöjdhet med livet hos föräldrar till barn med kronisk sjukdom.

Nyckelord: Föräldrar till barn med kronisk sjukdom, Mindfulness, KBT, Stress, Utmattning, HRQoL och Nöjdhet med livet.

**CONTENTS**

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## LIST OF PUBLICATIONS

I. Anclair M., Hovén E., Lannering B., & Boman K.K. (2009). Parental fears following their child's brain tumour diagnosis and treatment. *Journal of Pediatric Oncology Nursing 26*(2), 68–74.

II. Anclair, M., Lappalainen, R., Muotka, M., and Hiltunen, A.J. (in press). Cognitive Behavioural Therapy and Mindfulness for Stress and Burnout: A Waiting-List Controlled Pilot Study Comparing Treatments for Parents of Children with Chronic Conditions. *Scandinavian Journal of Caring Sciences.*

III. Anclair, M., Hjärthag, F. and Hiltunen, A.J. (2017). Cognitive Behavioural Therapy and Mindfulness for Health-Related Quality of Life: Comparing Treatments for Parents of Children with Chronic Conditions – A Pilot Feasibility Study, *Clinical Practice & Epidemiology in Mental Health 13*, 1–9.

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Hovén E., Anclair M., Samuelsson U., Kogner P., & Boman K.K. (2008). The influence of pediatric cancer diagnosis and illness complication factors on parental distress. *Journal of Pediatric Hematology/Oncology. 30*(11), 807–814.

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## LIST OF ABBREVIATIONS

ACT Acceptance and commitment therapy
ADHD Attention Deficit Hyperactivity Disorder
ALL Acute lymphoblastic leukaemia
AML Acute myeloid leukaemia
ANS Autonomic nervous system
ASD, Autism spectrum disorder
BP Bodily pain
CBI Cognitive behavioural intervention
CBT Cognitive behavioural therapy
CI Confidence interval
CNS Central nervous system
ED Exhaustion disorder
FOR Fear of progression/recurrence
FT Family therapy
GAS General adaptation syndrome
GH General health
HPA Hypothalamic-pituitary-adrenal axis
HRQoL Health-related quality of life
ICD-10 International Statistical Classification of Diseases and Related Health Problems,10th revision
MBI Mindfulness-based intervention
MBCT Mindfulness-based cognitive therapy
MBSR Mindfulness-based stress reduction
MCS Mental component summary
MH Mental health
MST Multisystemic therapy
PBSE Performance-based self-esteem
PCS Physical component summary
PE Physical function
PSS Perceived stress scale
PST Problem-solving therapy
PTSD Post traumatic stress disorder
QoL Quality of life
RCT Randomised control trials
RF Role functioning – emotional causes
RP Role functioning – physical causes

SD Standard deviations
SF Social functioning
SF-36 Short-form 36
SIT Stress inoculation training
SMBQ Shirom-Melamed Questionnaire
VT Vitality
WHO World Health Organization

## INTRODUCTION

Being a parent of a child who suffers from a chronic illness is a major challenge both psychologically and practically. When a child is diagnosed with a chronic illness or disability, it is a significant stressor that have an impact on both the child's and the parents' emotional and social functioning. The distress that arises can be more demanding than the illness itself. Parents are not only responsible for taking physical care of their child, but must also deal with the disease in terms of medical, school-related and other social aspects. The daily care of a child with a chronic disease or disability is demanding and can lead to increased and long-term burden, stress and fatigue (Appels & Schouten, 1991; Melamed, Kushnir, & Shirom, 1992; Strike & Steptoe, 2004; Toker, Shirom, Shapira, Berliner, & Melamed, 2005). Because medical treatments are becoming more effective for several serious diseases, the proportion of children with chronic conditions has increased as they live longer. Some chronic conditions in children, such as diabetes type 1 (T1D), have increased more than others during the recent decade (Berhan et al., 2011). The prevalence of children clinically diagnosed with a neuropsychiatric disorder has increased, whereas the prevalence of children with autism symptoms has remained stable (Lundström, Reichenberg, Anckarsäter, Lichtenstein, & Gillberg, 2015), as has the occurrence of childhood cancer (Asdahl et al., 2015). Consequently, there are numerous parents who suffer from stress-related mental illness and need targeted support for their own problems. Hopefully, in the near future, parents of children with chronic conditions will be offered targeted evidence-based interventions in regular healthcare, which will help them help themselves and their children become better equipped to meet demands and stressors.

### A biopsychosocial perspective on parental fears, stress and burnout

#### *Parental fears*

In this thesis, parental fears refer to the unique worries and uncertainty caused by illness and treatment that trouble parents of children with central nervous system (CNS) tumour and cancer (van Dongen-Melman et al., 1995). Worry involves directing attention to thoughts and images concerned with potentially negative events in the future (Borkovec, Robinson, Pruzinsky, & DePree, 1983). Worry is a type of attentional deployment, and worrying is generally considered maladaptive and a common feature of anxiety disorder (Borkovec & Inz, 1990).

### Stress definitions

The concept of stress can be defined in a number of ways. Some scientists believe that the concept is so worn out and vague that it should not be used (Jones, Bright, & Clow, 2001), while others defend the use of the term and make efforts to clarify and specify the stress concept (Cassidy, 2003; Lazarus & Folkman, 1984). The most commonly used definition of stress in Sweden is summarized by the Swedish National Board of Health and Welfare: 'Stress is the organism's reaction to the imbalance between the burden to which it is exposed to and the resources it has to deal with it' (Socialstyrelsen, 2003). From this definition of stress as an imbalance between burden and resources follows that not only a shortage, but also a substantial surplus, of resources against the strain that the organism is exposed to could result in some form of negative stress. The concept of stress thus includes both stressors and that which leads to stress responses. Stress responses, in turn, is a broad concept involving physiology, cognitions, emotions and behaviours.

### *The stress–vulnerability model*

Today, the stress–vulnerability model (Zubin & Spring, 1977) is well established and used in healthcare. "Vulnerability" refers to our basic susceptibility to mental health disorders. This is determined by our genetic predispositions and our early life experiences as well as by our emotions, thoughts and behaviours (Goh & Agius, 2010). The stress–vulnerability model emphasizes the interaction between stress and vulnerability: the higher the vulnerability, the lesser the stress needed to develop mental illness. If the stress is high enough, or prolonged, anyone can develop a mental or physical illness (Linton, 2013). According to this definition, stress-related problems are developed and maintained through interplay among biological, psychological and social environmental factors, often referred to as the biopsychosocial model of stress (Engel, 1977).

### *Distress*

Psychological distress is the unique, uncomfortable emotional state of a subject that occurs in response to a specific stressor or demand and results in injury – either temporarily or permanently (Ridner, 2004). Further, distress is also defined as a non-specific biological or emotional response to a demand or stressor that is stressful for the individual. The concept of distress can be seen as an inability to deal with what is perceived as a stressful condition. Psychological distress can be seen as a continuum where 'mental health' and

'mental illness' find themselves at opposite ends of a spectrum: from normal feelings about vulnerability and sadness, to the problems that can become disabling, such as depression, anxiety, panic, social isolation and existential crises. But also parental fears can cause distress. As we experience different things, we move on this continuum at different times throughout life (National Comprehensive Cancer Network, 2005). Factors described as strongly associated with distress include worries, uncertainty about the future, avoidance coping styles, escape and safety behaviours and difficulties in regulating emotions (Aldao, Gee, De Los Reyes, & Seager, 2016; Gross, 2014; Hetzel-Riggin & Meads, 2016; Pitceathly & Maguire, 2003; Utens et al., 2000).  Research has also shown that high levels of distress are associated with impaired quality of life (QoL) (Arafa, Zaher, El-Dowaty, & Moneeb, 2008; Davis, Davies, Waters, & Priest, 2008; Reilly, Taft, Nelander, Malmgren, & Olsson, 2015).

### Biological factors

*General Adaptation Syndrome (GAS)*

Hans Selye identified a general development of responses to stressful events in the form of physiological, psychological and behavioural responses known as GAS (Selye, 1950).

GAS is divided into three phases: alarm phase, resistance phase and exhaustion phase. In the alarm phase, a series of physiological changes happens. The acute stress or fear occurs when the individual tries to control challenging or threatening situations. The reaction is then usually intense but short-lived, and the stress hormones adrenaline and noradrenaline are secreted. The most instinctive response in this situation is the fight or flight response (see below).

In the resistance phase, the body strives to regain balance, homeostasis. If the resistance is successful, it will cease or reduce the effects of stress.  If the stressor is too extreme, the person may simply be unable to cope with it. Usually, the individual gathers his or her physical and/or emotional strength and begins to resist the negative effects of the stressor by using coping strategies or adapting to the environment.

If resistance fails, the exhaustion phase will follow. The body does not have enough energy to combat stress anymore. If the individual remains in a state of resigned stress for a long time, it leads to a lack of recovery – often with high secretion of the stress hormone cortisol. Frequent, acute stress reactions usually result in prolonged stress, which after many years can lead to fatigue and exhaustion reactions and to long-lasting stress-related health problems.

According to Selye, stress factors are called stressors, regardless of whether the threats/demands are external or internal. The stress concept is completely value-neutral: stress is neither positive nor negative but the body's defence to a threat (Selye, 1950).

*Fight, flight and freeze responses*

The body's autonomic nervous system (ANS) encompasses two systems of central importance to stress and recovery – the sympathetic and parasympathetic nervous systems – and one biological function known as the hypothalamic-pituitary-adrenal (HPA) axis. The general function of the ANS is to regulate physiological bodily functions such as digestion, body temperature and blood pressure (Almén, 2007; O'Connor, O'Halloran, & Shanahan, 2000). Acute stress activates our fight and flight response (sympathetic) through the stress hormones norepinephrine and epinephrine. All emotional reactions, such as anger, fear and anxiety, activate the sympathetic nervous system. The parasympathetic system is activated during relaxation and recovery, and hormones are released (e.g. oxytocin, which is referred to as the body's calming and relaxing hormone). Ongoing activation of the HPA axis and cortisol release occur during prolonged stress.

During chronic stress the HPA axis and the sympathetic nervous system become overactivated. Chronic stress appears to hamper the functions of the parasympathetic system, that is, when the individual tries to recover from it, anxiety and restlessness increase instead of calmness (Wahlberg et al., 2009). In cases of a perceived threat where the brain concludes that fight or flight is not possible, the body's freeze response can become activated. Also the freeze response is activated by the parasympathetic system. During lengthy, threatening situations, however, this braking system may become too dominant. It is this type of passive stress reaction that is most common for prolonged stress, and it triggers fatigue and need for comfort and recuperation. This type of stress reaction is also common for parents of children with chronic conditions who are affected by long-lasting parental fears, stress and burnout.

**Social and environmental factors**

Stressors can range from daily hassles to ongoing stressors. An individual's personal world can both be a source of stress and a source of support. Strenuous relationships in the family or in other social relationships can be sources of stress, whereas supportive relationships can assist in problem solving and help a person cope with life stressors (Cohen & Wills, 1985). Karasek and Theorell's

model is currently the prevailing model for understanding how psychosocial conditions affect our health (Karasek & Theorell, 1990). According to the model, it is the person's perceived demands and perceived control in a work situation that determines whether the work will lead to positive or negative stress. High external demand and low personal control can lead to a state of negative tension and eventually cause mental and physical ill health. Demands are defined as psychological stressors and stress factors (e.g. workload and time pressure), but can also be divided into physical and psychological demands. Control is defined by the degree of self-determination, stimulation and self-development (e.g. variation in work tasks). Recently, yet another factor was added to the model, social support. Social support has proved protective against stress, which means that persons with high social support have fewer stress symptoms than people who lack social support (Magnusson Hanson, Theorell, Oxenstierna, Hyde, & Westerlund, 2008; Karasek & Theorell, 1990). And social support for parents of children with chronic conditions has been shown to decrease distress (Stremler, Haddad, Pullenayegum, & Parshuram, 2017).

### Psychological factors

#### Transactional model of stress

In the psychological tradition, it is emphasized that stress occurs in the interaction between the individual and the environment. Stress arises when the individual's resources are exceeded and thus threaten the person's well-being. Today, the so-called transactional model of stress and coping, based on Lazarus and Folkman (1984), is one of the most established models in stress research (Cassidy, 2003). The model focuses on the individual's interpretations of the situation, and it is this process of interpretation that will determine how the individual will react (Cohen et al., 1995; Cohen, Tyrrell, & Smith, 1993). The interpretation process consists of two different cognitive evaluations (appraisals) divided into conscious or intuitive judgments. If the person is experiencing a situation as stressful, some kind of action is needed (Lazarus & Folkman, 1984). This means that the individual ways to manage stress and to cope with it become a central concept. According to Lazarus and Folkman (1984), coping means the cognitive and behavioural efforts a person make to manage external and internal demands. The transactional model of stress and coping is useful in modern psychological and medical science. Findings support that negative appraisals (i.e. harmful or threatening stimuli) are associated with negative psychological and physical adjustment, whereas positive appraisals

(i.e. challenges) are associated with positive psychological and physical adjustment. Appraising an event as catastrophic is associated with passive pain coping, venting, helplessness and increased levels of pain. At the extreme, these negative appraisals have been implicated in the development and maintenance of post-traumatic stress disorder (PTSD). Further, when a stressor is appraised as controllable, it is directly related to positive psychological adjustment. In addition, meaning-focused coping, which is an attempt to alter the meaning of a situation to make it more consistent with the individual's beliefs and goals, is also directly related to adjustment (Hauser-Cram et al., 2001; Larose & Bernier, 2001; Lau & Morse, 2001; Tunali & Power, 2002).

*Coping strategies*

The stress coping strategies have two main functions: regulate distress and take actions to deal with the source of distress (Lazarus & Folkman, 1984). There are different types of strategies to cope with stressful or threatening situations. There is *problem-focused coping,* where the focus is on solving the real problems that induce stress (e.g. systematic problem solving, knowledge about the situation, make up an action plan and follow it and seek help to reduce stress). *Emotion-focused coping* is about managing emotional reactions rather than focusing on the particular problem (e.g. relaxation, mindfulness, acceptance, perspective taking and exercising to reduce the negative effects of stress). *Appraisal-focused coping* emphasises cognitive assessment and logical analysis to increase confidence that the situation is manageable. Another important distinction prevalent in coping literature is that of active versus avoidant coping (Carver, Scheier, & Weintraub, 1989). *Active coping* involves exerting efforts of some kind to eliminate or minimise stressful events. In active coping individuals acknowledge the stressor and take measurable steps to resolve the issue. In contrast, *avoidant coping* relies on disengagement. This type of coping results in dismissal of, or an attempt to suppress, the problem. Common avoidant coping strategies are overconsumption of food and alcohol, and escape and safety behaviours. These general coping styles (problem and emotion-focused vs. active and avoidant) have specific strategies that overlap one another. Ultimately, the chosen coping mechanism depends on the individual, his/her resources and the type of stressor. Moreover, it is not uncommon for a combination of these mechanisms to be used for any given stressor (Lazarus and Folkman, 1984).

Research suggests that problem-focused coping and active coping are most often associated with positive outcomes and may also affect neurobiological

determinants (Bowen et al., 2014; Elumelu, Asuzu, & Akin-Odanye, 2015). Further, research shows that avoidant coping may be a modifiable predictor of mental illness such as sleep disturbance, poorer QoL, depressive symptoms and emotional problems and, in the long term, chronic stress development (Hetzel-Riggin & Meads, 2016; Myaskovsky et al., 2003; Taylor et al., 2015). Using active problem-focused coping and less avoidant behaviours are related to lower levels of anxiety and depression in all parents regardless of the health condition of the child (Luque Salas, Yanez Rodriguez, Tabernero Urbieta, & Cuadrado, 2017; Norberg, Lindblad, & Boman, 2005). Although the coping literature is somewhat inconsistent, there are no correct ways of coping. What coping efforts may work in one situation or at one time may not be applicable at other times (Compas, Forsythe, & Wagner, 1988; Kaloupek, White, & Wong, 1984). A more complete understanding of coping may require a microanalysis of coping processes in which individuals' flexibility to deploy different coping strategies in distinct stressful contexts is assessed (Cheng, 2001). However, a central concept underlying all stress management is that of bolstering an individual's repertoire of coping skills in a flexible fashion that meets the appraised demands of the stressful situation (O'Donohue & Fisher, 2008).

*Cognitive approach to emotion*

The study of emotions has expanded in psychology and extended to fields that range from history to neuroscience. There are currently intense debates on how to define emotions and how to best measure them (Oatley & Johnson-Laird, 2014). Cognitive theories of emotions based on the mind's organization of knowledge offer a clarifying perspective because they focus on the fundamental issues of how emotions are caused and what their effects are (Keltner, Oatley, & Jenkins, 2014). There are several cognitive theories of emotions, but they all have in common that emotions are caused by appraising events in relations to concerns (Frijda, 1988; Oatley & Johnson-Laird, 2014; Russell, 2003). The emotion is experienced as positive when the person reaches his or her goals, and as negative when this does not happen. The core of the emotion is to get us ready to act and launch plans. An emotion activates and accelerates one or more actions and can outmanoeuvre other mental processes such as thoughts and behaviour. A feeling is perceived as a distinct mental condition that can lead to bodily changes, linguistic activity and bodily expression.

One cognitive theory of emotions that is under development is Frijda's action readiness theory (Frijda, 2016). According to this theory, emotions are an assemblage of processes that contribute to how we relate to the social and

physical world, and the key is to understand the functional bases of such processes. One such process is action readiness. It is the preparation for a movement or action. The preparation for a movement occurs in all animals, and action readiness is the basis for the states we call emotions. Some basic emotions – happiness, sadness, anger and fear – can occur without an object. Fear is an important biological defence against a perceived threat. It affects the whole brain and the ANS, including the amygdala and neocortex, to alert the individual to danger or threat. The goal is to make us willing to eliminate the threat. Fear can be more or less functional depending on the circumstances. It makes no difference to fear whether the threat comes from outside or inside, is constructed or real (Keltner et al., 2014). The parental fears of parents of children with chronic conditions can also be understood in the light of Frijda's action readiness theory (Frijda, 2016) as parents prepare for a movement or action to reduce a threat. This theory is also similar to the GAS alarm phase and the fight, flight and freeze responses. Parental fears or worries can thus be seen as a stressor that affects the whole brain and can, if long-lasting, lead to chronic stress reactions and exhaustion.

*Emotion regulation*

Emotion regulation has been conceptualized as processes through which individuals modulate their emotions consciously and unconsciously (Gross, 1998b; Rottenberg & Gross, 2003). According to Gross (1998a), there are two types of emotion regulation: antecedent-focused emotion regulation and response-focused emotion regulation.

Antecedent-focused emotion regulation can determine whether emotional experience will happen and thus occurs before the emotion. For example, reappraisal, an antecedent-focused emotion-regulation strategy, occurs in the early emotion-generative process when emotional responses are not fully generated. Response-focused emotion regulation, by contrast, refers to the management of emotional impulses when emotions are already generated. Further, suppression, a response-focused emotion-regulation strategy, engages in the process of the behavioural modulation of emotional response, which occurs in the late emotion-generative process. Research shows that emotion regulation is increasingly incorporated into models of psychopathology as stress-related disorders (Conklin et al., 2015; Greenberg, 2002; Mennin & Farach, 2007). Several theorists argue that individuals who cannot effectively manage their emotional responses to everyday events experience longer and more severe periods of distress (Goldsmith, Chesney, Heath, & Barlow, 2013;

Mennin & Farach, 2007; Nolen-Hoeksema, Wisco, & Lyubomirsky, 2008). According to Gross (1998), attentional deployment involves directing one's attention towards or away from an emotional situation: Rumination, worry and thought suppression, for example, are all maladaptive emotion-regulation strategies associated with anxiety disorder and major depression (Borkovec & Inz, 1990; Nolen-Hoeksema et al., 2008).

Research has shown that appraisals are not just post hoc impressions, but causes of emotions and that the appraisal of the situation determines the emotion. The best way to reduce the intensity of a distressing emotion is by reappraising – generally considered to be an adaptive emotion-regulation strategy – both the events giving rise to it and the current situation (Siemer, Mauss, & Gross, 2007). By appraising emotions as unacceptable and suppressing them, the intensity of the emotion increases; the opposite, accepting emotions without judging, reduces distress (Campbell-Sills, Barlow, Brown, & Hofmann, 2006). Antecedent-focused emotion regulation, such as reappraisals and acceptance, is related to less distress (Almeida, 2005). Response-focused regulation is a less adaptive strategy to regulate emotions and is associated with greater sympathetic activation of the cardiovascular system (Gross, 2002). These findings are also supported by neuroimaging studies of cognitive emotion regulation (Ochsner & Gross, 2008) showing that reappraisal (early frontal engagement) leads to decreased amygdala/insula activity over time, whereas suppression (late frontal engagement) causes increased amygdala/insula activity over time. Further, cognitive emotion regulation (reappraisal) is not a unitary ability as it can be broken down into the subcomponents attention, response selection, working memory, language, mental-state attribution and autonomic control (Eysenck, Derakshan, Santos, & Calvo, 2007; Ochsner & Gross, 2008). These processes are also assumed to have sufficient empirical evidence to be transdiagnostic (Harvey, Watkins, Mansell, & Shafran, 2004). Today, several therapeutic approaches incorporate some form of emotion-regulation training, for example, emotion-focused therapy (Greenberg, 2002) and acceptance and mindfulness-based therapy (Gilbert, 2014a; Hayes, 2004; Segal, Williams, & Teasdale, 2013).

*Coping and emotion regulation*

The development of coping and emotion-regulation skills reflect the coordination and interplay of processes of social, cognitive, affective and brain development (Compas et al., 2014). Further, coping and emotion-regulation skills play a central role for a range of psychological problems and disorders in

transdiagnostic models of preventive interventions and psychological treatments (Compas et al., 2014). In contrast to coping, which is context dependent, emotion regulation involves the regulatory process of emotion in a broader sense (Wang & Saudino, 2011). Emotion regulation includes control of both positive and negative emotions and focuses primarily on the modulation of internal emotional changes so as to meet external needs. Coping not only involves internal emotion regulation, but also helps take control of external events. Accordingly, coping is broader than emotion regulation in that coping includes the adjusting process of both external problems and internal emotions, whereas emotion regulation mainly involves the regulation of inner whole-body responses. Hence, there is clear evidence that coping and emotion regulation are distinct but closely related constructs (Compas et al., 2014; Wang & Saudino, 2011).

*Coping, emotion regulation and psychological flexibility*

In a review article made by Kashdan and Rottenberg (2010), they conclude that psychological flexibility is an important factor for understanding psychological health. According to the review, psychological flexibility is a slippery construct to define. It comprises a wide range of human abilities such as to recognise and adapt to different situational demands; shift mindsets or behavioural repertoires when these strategies interfere with personal or social functioning; maintain balance among important life domains; and be aware, open, and committed to behaviours that are congruent with deeply held values (Kashdan & Rottenberg, 2010). In many forms of psychopathology, these flexibility processes are absent. Furthermore, research shows that there is a relationship between adaptive coping strategies and psychological flexibility (Cheng, Lau, & Chan, 2014). Flexibility in coping strategies and emotion regulation is related to better effectiveness in managing stressors, greater well-being and fewer symptoms of depression, anxiety and pain compared with inflexible coping strategies and difficulties in emotion-regulation (Cheng et al., 2014; Kashdan & Rottenberg, 2010; Wicksell, Olsson, & Hayes, 2010). Individuals with psychological flexibility may therefore be better equipped to consider and employ various coping and adaptive emotion-regulation strategies in order to resolve situations and reduce distress (Hayes, 2005).

**Burnout**

The term burnout was first used in a psychological-psychiatric context by Freudenberger (1974) to describe a condition that staff working with difficult

psychiatric patients showed. In the mid-1970s, American psychologist Christina Maslach (1978) claimed that a group of social workers showed a similar condition in their work with clients. Based on interviews, she identified three main dimensions of 'burnout' in staff in care-related professions: emotional exhaustion, loss of empathy towards clients and subjective deterioration of work performance (Maslach, 1978). Since 1981, these three dimensions have formed the core concept of burnout (Maslach & Jackson, 1981). However, the concept has widened slightly. Both physical and emotional exhaustion have been more emphasised and can be seen as a general exhaustion response due to long-lasting emotionally demanding situations in both professional and personal life (Pines & Aronson, 1988). The concept of burnout has many similarities with work-related stress. Much research describes the psychophysiological and behavioural stress responses with numerous references to burnout (Kahn & Byosiere, 1992). Shirom and Melamed (1989) defines burnout as a consequence of a prolonged stress reaction where the organism's resources to deal with difficulties become exhausted. In this approach, burnout relates to a condition where the organism is no longer able to restore balance in a prolonged stressful situation. This condition is popularly described as a 'stress collapse' or 'breakdown'.

Other common conditions that may be related to prolonged stress and fatigue are sleep and memory disorders, pain-related conditions, hypertension, type II diabetes, anxiety, depression and extreme tiredness that is not alleviated by rest (Danielsson et al., 2012). In spite of the typical clinical picture, chronic stress disorder or burnout is not yet recognized in any of the major psychiatric classification systems. In the International Statistical Classification of Diseases and Related Health Problems, 10th revision (ICD-10) (World Health Organisation, 1992), burnout is described as 'Problems related to life management difficulty' (Z73.0), not as a medical condition (Beser et al., 2014).

*Clinical burnout/Exhaustion disorder (ED)*

Exhaustion disorder (ED) is currently a medical diagnosis in Sweden. In 2005, the diagnosis was accepted and given the ICD-10 code F43.8 (Åsberg, Nygren, & Nager, 2013). The description of the syndrome largely corresponds to the core components of clinical burnout (Grossi, Perski, Osika, & Savic, 2015). ED is based on major specific diagnostic criteria – such as lack of psychological energy, cognitive deficits, reduced ability to cope with demands and/or time pressure, emotional instability, disturbed sleep and physical symptoms such as muscular pain – provided that the diagnosis of depression could be excluded.

Further, the symptoms need to be attributed to identifiable stressful events such as increased workload extending over a long period of time (> 6 months) (Åsberg et al., 2013).

ED differs from depression with respect to triggering causes, symptoms, biochemistry and treatment needs (Åsberg et al., 2013). Patients with ED have a reduced sensitivity of the HPA axis, which indicates that what happens in the body of someone with ED is different from depression. There is, for example, much evidence of increased sensitivity of the stress system during major depression. Moreover, research has shown that the reduced sensitivity of those who have, or have had, ED appears to be permanent, or at least remained reduced at follow-up after seven years (Åsberg et al., 2013).

The typical ED is generally preceded by a prodromal phase that can last for several years with fluctuating symptoms. At this stage, the symptoms are identifiable through interviews or assessment scales. The second stage of ED is an acute phase, often with dramatic symptoms, which usually subside within a few weeks, to be replaced by a recovery phase, which can last for many years (Åsberg et al., 2013).

The controlled treatment studies of different rehabilitation models during the recovery phase have not produced any convincing evidence that one specific approach would be better than another, with one exception: treatments that include the workplace appear to provide better results (Åsberg et al., 2013). A Swedish study shows that demands at work, relationship problems and caring for sick relatives or for children with disabilities are the most common self-reported causes of ED. Women more often reported private factors; men slightly more often reported work-related issues. The study concludes that it is the overall burden that is important and that it usually involves both private and working life (Hasselberg, Jonsdottir, Ellbin, & Skagert, 2014). In addition, research shows that people with ED often suffer from comorbidity with anxiety and depression (Glise, Ahlborg, & Jonsdottir, 2014). The literature also supports the notion that sleep impairments and insufficient recovery are causative and maintaining factors for this condition (Grossi et al., 2015; Söderström, Jeding, Ekstedt, Perski, & Åkerstedt, 2012).

### *In summary*

Parental fears, stress and burnout develop and are maintained through interplay among biological, psychological and social environmental factors, often referred to as the biopsychosocial model of stress (Engel, 1977). The stress-vulnerability model (Zubin & Spring, 1977) is well established and illustrates the interaction

between stress and vulnerability and shows that the outcome is greater than the separate parts (Goh & Agius, 2010). GAS is a term used to describe the body's short-term and long-term reactions to stress. Selye defines the three stages of the syndrome: alarm reaction, the stage of resistance and the stage of exhaustion (Selye, 1950). Frijda's action readiness theory (Frijda, 1988) is similar to both the GAS alarm phase and the fight, flight and freeze responses. Parental fears can thus be seen as a stressor that, if long-term, can lead to chronic stress reactions and exhaustion. The transactional model of stress (Lazarus & Folkman, 1984) emphasizes the interdependent relationship between the individual's functioning and environmental factors. This is in line with modern medical and psychological science, claiming that the individual is part of a context and must therefore be seen in the light of it (Hauser-Cram et al., 2001; Larose & Bernier, 2001; Lau & Morse, 2001; Tunali & Power, 2002). Emotion regulation is also increasingly incorporated into models of psychopathology (Conklin et al., 2015; Greenberg, 2002; Mennin & Farach, 2007). Several theorists argue that individuals who cannot effectively manage their emotional responses to everyday events experience longer and more severe periods of distress (Goldsmith et al., 2013; Mennin & Farach, 2007; Nolen-Hoeksema et al., 2008).

Coping is broader than emotion regulation in that coping includes the adjusting processes of both external problems and internal emotions, whereas emotion regulation mainly involves the regulation of inner whole-body responses. Regardless of the differences between emotion regulation and coping, it is clear that these two concepts overlap considerably (Wang & Saudino, 2011). The relationship between emotion regulation and stress can also be explained by the common neural structures, including prefrontal cortex, anterior cingulate cortex and amygdala, which are all engaged in the process of emotion regulation and stress response (Ochsner & Gross, 2008). Karasek and Theorell's model (Karasek & Theorell, 1990) further illustrates that it is an individual's total imbalance between demands and resources and between stress and recovery that is important. Parental fears, stress and burnout symptoms are expressed in thoughts, feelings, behaviours and physiological responses. Examples of these are worries, catastrophizing, rumination, anxiety, escape and avoidance behaviour, passivity, fatigue, decreased immune function, hypertension, metabolic syndrome and insomnia (Pitceathly & Maguire, 2003; Utens et al., 2000). These symptoms are all expressions of distress, often leading to impaired QoL (Arafa et al., 2008; Davis et al., 2008; Reilly et al., 2015). After several years, vulnerability, together with lack of resources, maladaptive coping

strategies (Luque Salas et al., 2017; Norberg et al., 2005) and difficulties in emotion-regulation (Goldsmith et al., 2013; Mennin & Farach, 2007), can result in long-lasting distress and burnout in parents of children with chronic conditions. The biopsychosocial stress model thus offers a comprehensive theoretical understanding of stress as well as three major foci for the assessment and treatment of a variety of stress-related disorders (O'Donohue & Fisher, 2008), including parental fears, stress and burnout in parents of children with chronic conditions.

**Parents of children with chronic conditions**

### *Chronic disease and health condition*

The most common definition of chronic conditions in children is based on the ICD-10 classification of the World Health Organisation (1992). A condition can be considered chronic if (1) it occurs in children aged 0 to 18 years; (2) the diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods or instruments according to professional standards; (3) it is not (yet) curable or, for mental health conditions, it is highly resistant to treatment and (4) it has been present for longer than three months or will probably last longer than three months, or it has occurred three times or more during the past year and will probably reoccur. The most common chronic conditions can be divided into the following groups: cardiovascular disease, cancer, diabetes, lung diseases, mental illness, neurological disorders, muscle and joint disorders and chronic pain (Martin, 2007). A disability is an impairment of physical, mental or intellectual functioning. A disability can occur as a result of illness or other conditions, or as a result of a congenital or acquired injury. Such diseases, conditions or injuries can be permanent or transitory (Darcy et al., 2015).

It is difficult to get an overview of how many children there are who meet the criteria for chronic conditions in Sweden today. It is not easy to determine how serious these chronic conditions are when, for example, both uncomplicated allergies and cancer count as chronic conditions. Some children also suffer from several serious chronic conditions at the same time. A clearer division of severe, moderate and mild conditions would therefore facilitate understanding. Even the definition of chronic disease varies in studies, which further complicates the picture. However, a study from the 1980s (Westbom & Kornfält, 1987) examined the incidence of chronic disease in an area with ten schools in southern Sweden. All children's health records were examined. Chronic conditions were revealed

in 510 out of the total population comprising 6080 children less than 16 years of age. The study found that 8% of the chronically ill children met the criteria for severe disability (mental and nervous system disorder and congenital malformations); 22% met the criteria for moderate disability (T1D, epilepsy and speech disorder); and 70% met the mild disability criteria (diseases of the respiratory system, otitis media and enuresis/incontinence) (Westbom & Kornfält, 1987). Another Swedish survey study from 2011 showed that almost 25% of the 2510 children aged 10, 12 and 15 years answered that they had some form of chronic condition such as T1D, attention deficit hyperactivity disorder (ADHD), mental disorder, hearing/vision/physical impairment or allergy (Svensson, Bornehag, & Janson, 2011). A review of studies from the Nordic countries, Israel and the United States showed that the lowest rate of prevalence of disabilities in children was found in the United States (5.8%) and the highest in Finland (9.8%) (Merrick & Carmeli, 2003). In 2007, a US study showed that the prevalence of chronic disease in children tripled over a 20-year period: Today, approximately 7% of all American children meet the criteria for chronic disease (Perrin, Bloom, & Gortmaker, 2007). Hence, there seems to be an increase in the number of children surviving childhood with diseases or conditions that once were considered life-threatening.

*Parents of children with central nervous system (CNS) tumour, cancer or traumatic brain injury*

As a result of advances in diagnostics and treatment during past decades, childhood cancer has evolved from a once acute fatal illness to what has been compared to a chronic condition (Fuemmeler, Elkin, & Mullins, 2002). In Sweden, about 300 children are diagnosed with cancer each year. Today, almost 80% of the children and adolescents diagnosed with cancer become long-term survivors, although the survival rate differs depending on the type of cancer (Asdahl et al., 2015). In developed countries, CNS tumours represent the largest group of solid tumours of childhood. In the Nordic countries, they constitute about 28% of all paediatric malignancies with an incidence of 4.2:100 000/year (NOPHO, 2008). Malignant tumours are the third most frequent cause of death among children aged 1–14 years, following congenital malformations and accidents (Gatta et al., 2014). Survival depends on both diagnostic and prognostic factors. Leukaemia is a type of cancer that arises in the bone marrow. There are different types of leukaemia, but the most common ones are acute lymphocytic leukaemia (ALL) and acute myeloid leukaemia (AML). Approximately 86% of the children suffering from ALL survive, while the

prognosis for AML is about 60% (Gatta et al., 2014). However, despite advances in treatment, childhood cancer remains the most common cause of childhood disease-caused deaths in the Western countries (Kaatsch, 2010; Pritchard-Jones, Kaatsch, Steliarova-Foucher, Stiller, & Coebergh, 2006).

The situation of parents after a cancer diagnosis is characterised by the need for general information about the illness, but also by the need for individual answers to unique questions. Research on parents of children with chronic conditions has identified the following frequent psychological effects: deteriorating life quality, stress-related disorders, PTSD, compulsive thought patterns, evasion, insecurity, fears and despondency (Sultan, Leclair, Rondeau, Burns, & Abate, 2016; Whalen, Odgers, Reed, & Henker, 2011). The initial reactions of parents of children with cancer have frequently been addressed in the literature, and anxiety and depression appear to be the most common ones (Boman, Lindahl, & Björk, 2003; Maurice-Stam, Oort, Last, & Grootenhuis, 2008; Sultan et al., 2016). Childhood cancer is characterized by a relatively great uncertainty not only about treatment success and prognosis, but also regarding late effects of treatment (Kaneko, 2009). Several studies have confirmed that children treated for CNS tumours are at increased risk for a variety of severe health-related, psychological and social long-term sequelae compared to other childhood malignancies (Hjern, Lindblad, & Boman, 2007; Johannesen, Langmark, Wesenberg, & Lote, 2007; Mulhern & Palmer, 2003). These studies reflect the fact that most children who survive experience some kind of adverse long-term consequences following the tumour and/or its treatment. Studies in this area show that impairments in attention, memory, learning capacity, language, and executive function are particularly common (Mulhern et al., 2005; Patel et al., 2014). Moreover, a decline in IQ over time is common in this population (Fouladi et al., 2005; Mulhern et al., 2005). The neurocognitive late effects range from mild learning issues to severe deficits in intellectual function (Mulhern, Merchant, Gajjar, Reddick, & Kun, 2004).

For parents of children with cancer, some risk factors, such as the number of the child's hospital admissions, may be significant predictors for parents' distress, especially for fathers (Sloper, 2000). Parent–child interactive stress may be influenced by variables related to 'child characteristics', 'parent characteristics' and 'environmental characteristics' (Mash & Johnston, 1990). A review investigating factors of distress in parents of children with cancer concludes that sex of the parent, coping response and personal resources, and pre-diagnosis family functioning were the major contributing reasons for distress, whereas education, income or marital status could not be attributed to differences in

distress (Sultan et al., 2016). For parents of children diagnosed with a brain tumour, the parents' perceived influence of the disease on everyday life predicted burnout symptoms – including subsymptoms of emotional exhaustion, fatigue, tension and cognitive difficulties (Norberg, 2010). Avoiding reminders of stressful experiences related to a child's cancer disease during and immediately after treatment seems to increase the risk of parents – mothers and fathers alike – later experiencing symptoms of post-traumatic stress (Lindahl Norberg, Poder, & von Essen, 2011). Moreover, restrictions in leisure activities during and after treatment is associated with PTSD symptoms in parents of children with cancer (Hovén, Grönqvist, Poder, von Essen, & Lindahl Norberg, 2017).

Fear of progression and fear of recurrence (FOR) of illness are appropriate and rational responses to real threats such as cancer and cancer treatment. Research has shown that FOR is one of the most common symptoms of distress in individuals with cancer or other chronic diseases (Berg et al., 2011). FOR is also common among parents of children with cancer or other chronic conditions (Fidika, Herle, Herschbach, & Goldbeck, 2015; Schepper et al., 2015). Elevated levels of FOR can increase psychological distress and affect well-being, QoL and social functioning. Recent systematic research suggests that about 50% of all cancer patients experience moderate to severe FOR (Herschbach & Dinkel, 2014). Parents of children with complicated cancer (CNS tumour and bone tumour) showed significantly heightened disease-related fear, anxiety, depression, loss of control, late effects-related uncertainty and poorer self-esteem compared with parents of children with ALL (Hovén, Anclair, Samuelsson, Kogner, & Boman, 2008).

Studies of families with children suffering from severe traumatic brain injury have shown that parents are vulnerable for experiencing persistent stress due to the child's injury (Aitken et al., 2009; Wade et al., 2006) and that caregiver distress increases as a function of time from the time of injury (Brooks, Campsie, Symington, Beattie, & McKinlay, 1986). Such persistent stress may result in chronic stress reactions or ED and lack of QoL.

*Parents of children with type 1 diabetes (T1D)*

During the past few decades, a rapid increase in childhood T1D has been reported from many parts of the world. Second to Finland, Sweden has the highest reported nationwide annual incidence of T1D in the world (Berhan et al., 2011). From 1978 to 1997, the incidence of T1D among children aged 0−15 years almost doubled in Sweden, with the largest increase among those aged 0−5

years. In Sweden, about 800 children are diagnosed with T1D each year (Berhan et al., 2011).

One Swedish study based on 252 parents of children with T1D showed that 36% of the parents suffered from clinical burnout compared to 20% of the parents of healthy children. The study found some psychosocial background factors that were significantly associated with burnout in parents of children with T1D; for example, low social support, lack of leisure time, the perception that the child's disease affects everyday life, low self-esteem and high need for control were risk factors for burnout (Lindström, Åman, & Norberg, 2010). Another study shows that in paediatric diabetes, the persistence or intensification of parental distress increases over time (Boman, Viksten, Kogner, & Samuelsson, 2004).

*Parents of children with mental health problems, ADHD, Autism spectrum disorder (ASD) and/or intellectual disability*

ASD is characterized by pervasive deficiencies in social interaction and communication and impaired imagination, which affect imaginative ability, behaviour and interests. The limitations must be sufficiently serious to affect the everyday life of children and the symptoms must be proved before the age of 3 years to be diagnosed (American Psychiatric Association, 2013). Autism is a lifelong disability that cannot be cured. Early intervention is therefore of great importance for promoting child development and increase functioning. Many children diagnosed with ASD also suffer from mental retardation (Hedvall et al., 2014). Comorbidity with other mental disorders is also very common: About 90% of all children with ASD meet the criteria of ADHD or one of the most common anxiety disorders such as generalized anxiety disorder or phobia (Salazar et al., 2015).

ADHD is characterized by difficulties with attention, impulse control and hyperactivity. The symptoms may occur singly or in combination (American Psychiatric Association, 2013). ADHD is a common disability: About 5% of all children have ADHD and problems often remain in adult life (Gillberg et al., 2004). It is also common that children with ADHD have other psychiatric problems; for example, both oppositional defiant disorder and conduct disorder as well as depression and anxiety disorders are common. Further, ADHD is highly heritable and multifactorial; multiple genes and non-inherited factors contribute to the disorder. Prenatal and perinatal factors have been implicated as risks, but definite causes remain unknown (Gillberg et al., 2004; Thapar & Cooper, 2015).

Several studies investigating variables associated with distress among parents of children with mental health problems showed that both internalizing and externalizing behaviour problems and psychosocial problems in children were significantly associated with parents' subjective and objective distress. Perceived personal control moderated the relationship between internalising child behaviours and parental subjective distress (Bussing et al., 2003; Duchovic, Gerkensmeyer, & Wu, 2009; Timko, Stovel, & Moos, 1992). Further, the child's behavioural and emotional impairments seem to predict the overall levels of distress (i.e. stress/tension, anxiety and depression) in parents of children with autism spectrum disorder (Firth & Dryer, 2013). A combination of child and parent demographics, severity of child behavioural disturbance, low knowledge of ADHD, causal and controllability attributions internal to the child, along with lower perceived parental control, are associated with more severe psychological distress in mothers of children with ADHD (Harrison & Sofronoff, 2002). The rate of ADHD and maternal stress is significantly higher in the group of children with lower interest in play (Weber-Borgmann, Burdach, Barchfeld, & Wurmser, 2014), and more ADHD symptoms are associated with poorer HRQoL in mothers (Coghill & Hodgkins, 2016). Another study showed that psychological adjustment to chronic stress, such as gastrointestinal symptoms, pseudoneurological symptoms and flu symptoms, were frequent –especially in mothers of children with intellectual disability (Vukojevic, Grbavac, Petrov, & Kordic, 2012).

*Parental distress and its relation to mental health problems in children*

Emotional and behavioural problems in children with chronic conditions are common. Research suggests that parenting has an important role to play in helping children become well-adjusted, and that the first few months and years are especially important (Barlow, Bergman, Kornor, Wei, & Bennett, 2016). Studies show that parental psychological distress increases the likelihood of mental health problems in their children (Amrock & Weitzman, 2014). In parents of both genders, associations existed between parental psychological distress and abnormal emotional symptoms in younger children, conduct disorder in older children and hyperactivity in children of all ages (Amrock & Weitzman, 2014). Research also shows that there is an association between parental distress, interaction with the child and child outcomes – such as glycaemic control and QoL – in children diagnosed with T1D (Nieuwesteeg et al., 2016). Another important finding is that chronic conditions in children

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increase the risk for physical abuse by their parents with 88% compared to children without chronic conditions (Svensson et al., 2011).

### *In summary*

There is strong scientific support that parents of children with chronic conditions suffer from distress. There are also numerous studies showing that parents of children with chronic illness have poor QoL (Arafa et al., 2008; Cappe, Bolduc, Rouge, Saiag, & Delorme, 2016; Reilly et al., 2015). It is therefore likely that the long-term stress of these parents will result in some form of chronic stress reaction, such as clinical burnout/ED; hence, stress may have serious negative consequences (Appels & Schouten, 1991; Melamed et al., 1992; Strike & Steptoe, 2004; Toker et al., 2005). The existing challenges that parents of children with chronic conditions struggle with are documented in studies of parents of children with, for example, cancer, T1D, chronic pain, ADHD, asthma, heart disease, CNS tumour, autism, schizophrenia, irritable bowel syndrome or arthritis (Boman et al., 2004; Lindahl Norberg, 2007; Lindström, Åman, & Norberg, 2011; Sullivan-Bolyai, Rosenberg, & Bayard, 2006; Wolf, Noh, Fisman, & Speechley, 1989). Moreover, almost all studies indicate that mothers of children with chronic conditions suffer from psychological distress and burnout to a greater extent than fathers (Lindström et al., 2010; Sultan et al., 2016; Weber-Borgmann et al., 2014; Yeh, 2002). Parents' distress may also have an impact on the children's mental health problems, and chronic conditions in children increase the risk for physical abuse by their parents (Svensson et al., 2011). It is therefore of utmost importance that these parents are offered adequate support. But it is equally important that they are good parents and support their child's development.

## Psychological interventions targeted at parents of children with chronic conditions

### *Cognitive Behavioural Therapy (CBT)*

CBT is an approach to human problems that can be viewed from several interrelated perspectives: philosophical, theoretical, methodological, assessment-oriented and technological (O'Donohue & Fisher, 2008). CBT is based on the psychology of learning and behaviour principles (Skinner, 1953), cognitive psychology (Beck, 1970) and social psychology (Bandura, 1989). It rests on around 20 different theories and is a combination of behavioural therapy and cognitive therapy. Further, CBT is an empirically validated form of

psychotherapy whose effectiveness has been proven in over 350 outcome studies of various mental disorders (e.g. depression, anxiety and eating disorders) and of stress coping and relationship problems treated individually and in groups (Butler, Chapman, Forman, & Beck, 2006; Öst, 2008; Roth & Fonagy, 2005). One review provides a comprehensive survey of meta-analyses examining the efficacy of CBT. The researchers reviewed a representative sample of 106 meta-analyses examining CBT for substance use disorders, schizophrenia and other psychotic disorders, depression and dysthymia, bipolar disorder, anxiety disorders, somatoform disorders, eating disorders, insomnia, personality disorders, anger and aggression, criminal behaviours, general stress, distress due to general medical conditions, chronic pain and fatigue, and distress related to pregnancy complications and female hormonal conditions. The review elucidates that the strongest support for CBT was found for anxiety disorders, somatoform disorders, bulimia, anger control problems and general stress (Hofmann, Asnaani, Vonk, Sawyer, & Fang, 2012).

CBT is a treatment where the therapist helps the client move towards new insights, re-learning and corrected emotional experience without interpretation, condemnation, or preconceived ideas. Cognitive restructuring, exposure, discriminative learning, identification of triggers/consequence analysis, problem-solving, validation, regulation of emotions, behaviour experiments and behaviour activation are common CBT techniques (O'Donohue & Fisher, 2008). In the 1980s, a form of cognitive behavioural intervention (CBI) called stress inoculation training (SIT) was developed by Meichenbaum (Meichenbaum & Novaco, 1985). SIT is a broad-based cognitive-behavioural intervention that provides a set of procedural guidelines to be individually tailored to the needs and characteristics of each client as well as to the specific form of stress that is being experienced. SIT includes three phases: a conceptual-educational phase; a skills acquisition, consolidation and rehearsal phase; and an application and follow-through phase. SIT has been used in varied populations, both on a preventive basis and on a treatment basis. Stress management techniques also derive from CBI and, together with SIT, teach clients new skills to help them cope with stress. Clients learn to use behaviours and thoughts for coping during stressful situations. They also commit to making life style changes that will lead to better QoL. Most stress management techniques based on CBI include psychoeducation about the biopsychosocial model of stress, coping skills (e.g. relaxation and coping thoughts) and lifestyle changes meant to decrease stress and increase QoL. These lifestyle changes should be consistent with the clients' interests and motivation. In addition,

clients are taught the ABC model of thinking and emotion (Ellis, 1973): How one's thoughts or beliefs (B) related to a situation or antecedent (A) can affect one's mood as a consequence (C). Moreover, psychoeducation often addresses common cognitive distortions and irrational beliefs that may impact clients' emotional reactions to stressful events (Beck, 1964).

### Mindfulness

Mindfulness means "maintaining a moment-by-moment awareness of our thoughts, feelings, bodily sensations and surrounding environment. Mindfulness also involves acceptance, meaning that we pay attention to our thoughts and feelings without judging them. For instance, without believing that there's a 'right' or 'wrong' way of thinking or feeling at a given moment. When we practice mindfulness, our thoughts tune into what we are sensing in the present moment rather than rehashing the past or imagining the future" (Kabat-Zinn, 2017).

More than thirty years ago, Jon Kabat-Zinn at the University of Massachusetts Medical Center in Worcester introduced a mindfulness-based training programme (Kabat-Zinn, 1982). In recent years, psychological research has paid increasing attention to mindfulness and its effect on mental health. Mindfulness-Based Stress Reduction (MBSR) and Mindfulness-Based Cognitive Therapy (MBCT) have proven to be effective methods for reducing distress, anxiety, depression and other mental conditions in a broad population (Benn, Akiva, Arel, & Roeser, 2012; Marchand, 2012; Martin-Asuero & Garcia-Banda, 2010; Segal et al., 2013; Teasdale et al., 2000).

One meta-analysis studied (Goyal et al., 2014) 47 randomised controlled trials of the effect of meditation-based treatment for various psychiatric disorders (e.g. anxiety, depression, substance abuse, eating disorders, pain syndromes and sleep disorders). The programmes studied were, among others, MBSR and MBCT. Effect sizes for anxiety, depression and pain syndrome were 0.38, 0.30 and 0.33, respectively. No greater effect was seen when compared with active control (Goyal et al., 2014). As a comparison, the effect size for drugs used for common illnesses has about the same effect size, 0.37; the effect size for drugs used in psychiatry is 0.41 (Leucht, Hierl, Kissling, Dold, & Davis, 2012).

Broadly, the goal of mindfulness training is to learn to observe a greater variety of experiences and sensations; to describe thoughts and feelings without judging or wishing they were different (antecedent emotion regulation, non-avoidance coping); to increase psychological flexibility; to learn to let the negative and

stressful thoughts come and go; and to be able to act consciously and effectively, while maintaining the capacity for self-compassion, which in turn results in positive psychological outcomes (Kabat-Zinn, 1982; Segal et al., 2013). A systematic meta-analysis identified moderate and consistent evidence for mindfulness, rumination and worry, and preliminary but insufficient evidence for cognitive and emotional reactivity, self-compassion and psychological flexibility as mechanisms underlying MBSR and MBCT (Gu, Strauss, Bond, & Cavanagh, 2015, 2016).

### *Previous research – CBT and Mindfulness for stress-related disorders*

A meta-analysis of the efficacy of CBT in treating chronic fatigue syndrome, which resembles the exhaustion disorder, included a total of 1,371 participants and showed a significant difference in post-treatment between participants receiving CBT and those in control conditions. Results indicate that CBT for chronic fatigue syndrome tends to be moderately efficacious (Malouff, Thorsteinsson, Rooke, Bhullar, & Schutte, 2008). Another study showed that symptoms decreased regardless of treatment, CBT or treatment as usual (de Vente, Kamphuis, Emmelkamp, & Blonk, 2008). Several meta-analyses have shown that mindfulness-based interventions (MBI) reduce stress-related mental illness and increase QoL (Khoury, Sharma, Rush, & Fournier, 2015). Another systematic review of a total of 209 studies concludes that MBI is a moderately effective treatment for a variety of psychological problems. MBI is especially effective for reducing anxiety and depression, and did not differ from CBT or behavioural therapies in pre–post comparisons (Khoury et al., 2013).

### *Psychological interventions aimed at parents of children with chronic conditions*

Psychological interventions are defined as psychotherapeutic treatment specifically designed to change parent cognition or behaviour, or both, with the intention of improving child outcomes (Eccleston, Fisher, Law, Bartlett, & Palermo, 2015). These interventions have developed over the years for parents of children with chronic illness, and have had a different focus. Some have been directed exclusively to parents; others have been targeted to both parents and children. The aim of these interventions has been to improve the health of both parents, children and other family members (Eccleston et al., 2015; Eccleston, Palermo, Fisher, & Law, 2012). The Cochrane review (Eccleston et al., 2015; Eccleston et al., 2012) is based on 35 randomized controlled trials (RCTs) involving a total of 2,723 primary participants consisting of parents of children

and adolescents (under 19 years of age) with a chronic illness, who were compared to an active control group. The chronic conditions that the children suffered from were painful conditions, cancer, T1D, asthma, traumatic brain injury, inflammatory bowel diseases, skin diseases or gynaecological disorders. The duration of treatment in the analysed studies varied between 3–16 hours. Data were analysed for each medical condition across all treatment classes at two time points (immediately post-treatment and the first available follow-up) and by each treatment class – CBT, family therapy (FT), problem solving therapy (PST) and multisystemic therapy (MST) – across all medical conditions at two time points (immediately post-treatment and the first available follow-up). The main results of this review show that across all treatment types, psychological therapies that included parents significantly improved child symptoms for painful conditions immediately post-treatment (Eccleston et al., 2012). Across all medical conditions, CBT significantly improved child symptoms and PST (in which cognitive behavioural strategies are used) significantly improved parent behaviour and parent mental health immediately post-treatment. All effects were immediately post-treatment. There were no significant findings for any treatment effects for any condition at follow-up.

One small Swedish study evaluated the effect of a group intervention by measuring changes in self-rated clinical burnout and performance-based self-esteem (PBSE). All parents who exhibited clinical burnout symptoms in accordance with the Shirom-Melamed Burnout Questionnaire (SMBQ) were then invited to participate in a group intervention. The group intervention consisted of eight sessions over a 12-week period and included education about behaviour, cognition and symptoms associated with burnout. The purpose of the intervention was to help the parents develop adequate strategies for coping with and reducing stress. SMBQ ($p = 0.01$) and PBSE scale ($p = 0.04$) measurements were significantly reduced, and the effects remained 6 months after completion of the intervention (Lindström, Åman, Anderzen-Carlsson, & Lindahl Norberg, 2016).

Forty-four parents, mostly mothers, of children with chronic conditions participated in a group MBSR study. Prior to the intervention, caregivers reported very high levels of stress and mood disturbance. Symptoms decreased substantially over the 8-week programme, with an overall reduction in stress symptoms of 32% and in total mood disturbance of 56% (Minor, Carlson, Mackenzie, Zernicke, & Jones, 2006).

### *In summary*

There is convincing evidence that CBI and Mindfulness are effective methods for reducing stress-related mental illness in general (Hofmann et al., 2012). Nonetheless, more work is needed to develop and provide psychological interventions that directly target parents of children with chronic conditions. Few interventions have provided intensive treatment that specifically target parent outcomes. The Cochrane review (2012) shows that CBT significantly improved child symptoms across all medical conditions, and PST significantly improved parent behaviour and parents' mental health immediately post-treatment. Further, the Cochrane review suggests that interventions targeting specific strategies aimed at parents' mental health and problem solving skills training are more likely to achieve those effects than interventions that include parents but do not purposefully target strategies.

## THE EMPIRICAL INVESTIGATIONS

### General aim

The general aim of the present research was to survey parental fears and investigate HRQoL, stress and burnout in parents of children with chronic conditions who were treated with mindfulness-based therapy or CBT.

### Specific aims

#### Study I

The aim of Study I was to investigate the fears that characterize parents after the CNS tumour diagnosis of their child, in order to advance our knowledge about how psychosocial support and information to these parents could be enhanced. A reference group of parents of childhood ALL patients, known to be at less risk for severe late effects of illness and treatment (Hjern et al., 2007), was used to specifically delineate the unique fears that characterize parents of children treated for CNS tumour.

#### Study II

The aim of Study II was to investigate if CBT or Mindfulness group treatment could reduce stress and burnout in a population of parents of children with chronic conditions.

#### Study III

The aim of Study III was to examine health-related quality of life (HRQoL) and life satisfaction of parents of children with chronic conditions before and after completed CBT or mindfulness intervention.

### Methods

#### Respondents and procedures, Study I

Study I included 82 parents of 48 children previously or presently treated for CNS tumours and 208 reference parents of 121 children previously or presently treated for ALL. Both parents received a questionnaire and instructions to complete without consulting each other. The data were collected as part of a larger research project investigating the psychosocial situation for parents of

children with cancer. The data collection of this larger study was made between October 2000 and April 2003.

Parents in Study I were consecutively recruited at two Swedish childhood cancer centres: Astrid Lindgren Children's Hospital and Linköping University Hospital. To meet the criteria for inclusion, parents had to have sufficient knowledge of the Swedish language to be able to comprehend the questionnaire. Knowledge in Swedish was considered insufficient if parents used an interpreter for communication with medical staff. Secondly, parents of children undergoing palliative treatment and parents who had lost their child were not approached; neither were parents of children with a known poor prognosis at the time, that is, parents of children for whom curative treatment had been resigned. Only cases of pontine glioma, known to be 100% incurable at the time of the study, were excluded *a priori* on the basis of diagnosis.

### Respondents and procedures, Studies II and III

Parents of children with chronic conditions suffering from stress and burnout symptoms were recruited to the project via advertisement in the local press in Karlstad, Värmland, Sweden. Parents whose child was under 18 years of age were invited to participate in the study. For participation, an average of more than 2.75 on the SMBQ scale was required and/or above 25 on the Perceived Stress Scale (PSS). All others were excluded from the study as they did not meet the stress and/or burnout criteria.

Parents with insufficient knowledge of Swedish (i.e. in need of an interpreter) to fill out the questionnaires were not invited to participate in the study. Parents undergoing other psychological treatment were also excluded.

Twenty-eight participants were initially recruited, but only 21 were included in the analysis, except for Study II, where 28 participants took part in the baseline phase of the study. A total of seven persons (25%) dropped out before the completion of the study. Seven clients withdrew from treatment in the initial phase and two in the treatment phase.

The participants' ages ranged from 36–54 years (CBT, 36–45 years, mean 40.4; Mindfulness, 41–54 years, mean 46.3). In all, 17 of the 19 participants were women: 9 out of 10 for the CBT group, 8 out of 9 for the Mindfulness group. Twelve of them (5 in CBT and 7 in Mindfulness) had more than 4 terms of academic education. The CBT group's children were 1–15 years old, with a mean age of 9.9 years; the Mindfulness group's children were 6–18 years of age, with a mean age of 11.4 years. The children's types of diagnoses were distributed as follows for Somatic illness/Psychiatric illness/Both: 1/6/3 (CBT) and 2/6/1

(Mindfulness). Each participant was informed about the study orally and in writing and gave their written consent. They were also informed that participation was voluntary and that they could withdraw from the study at any time (Table 1, Study II).

The study started with a baseline period ($n$ = 28). The baseline (waiting-list) period lasted for six months before they were randomized into one of the two treatment groups: Mindfulness (Mindfulness, ($n$ = 9) or CBT ($n$ = 10), (Figure 1, Study II). The purpose of the baseline period was to investigate the effect of measurements as well as the impact of participation and attention on measures of stress and burnout.

### Assessments, Study I

The list of parental fears in the questionnaire used was based on a categorisation made by van Dongen-Melman (1995). It covered particular cancer-related worries of parents of children with cancer. The conceptual framework of the assessment model of this original Dutch questionnaire is based on literature and in-depth interviews with parents of childhood cancer patients. The Swedish version of the questionnaire used here includes 11 items characterizing parental fears (Table 2, Study I). For each item, parents were asked to rate the intensity of fear along a 4-point Likert-type scale with response alternatives 1 (*not at all*), 2 (*a little*), 3 (*fairly*), and 4 (*extremely*).

### Assessments, Studies II and III

The parents filled out a questionnaire at the time of inclusion (6 months before the intervention), at the start of the intervention, and at the end of the intervention to measure the degree of perceived stress and burnout. Stress and burnout were measured with the SMBQ and the Perceived Stress Scale (PSS).

Depending on how the stress concept is defined, the procedures for measuring stress will differ. In cases where concern for the individual's subjective experience and contextual factors are of interest, the self-report instrument is a regular method of collecting data. The development of instruments assessing stress has gone from studying the actual stressors with instruments that measure dramatic life events, such as death and divorce, to instruments that focus on smaller, more frequent events in life, called hassles and uplifts (Kanner, Coyne, Schaefer, & Lazarus, 1981). However, Lazarus (1990) points out that the important thing is not whether the actual life event is small or large – it is how the event is valued by the individual that determines if it is a hassle or not. It is also important to take into consideration that persons living under more chronic

stressors (e.g. caring for a sick relative) may not be captured by hassles and uplifts instruments, as their experiences are not perceived as something extraordinary (Lepore, Palsane, & Evans, 1991). Hence, there appears to be a substantial overlap between the concepts of chronic stressors, life events and hassles in literature (Hahn & Smith, 1999).

Another way of measuring stress is to measure the actual symptoms thought to be the result of stress, which is usually referred to as output instead of input (Lazarus, 1990). Some instruments measure different symptoms usually associated with stress; others measure the individual's subjective experience of stress. However, both instrument types aim to capture the core concept of stress. One of the most used instruments that measures the individual's perception of stress is the PSS (Cohen, Kamarck, & Mermelstein, 1983).

*The Perceived Stress Scale, (PSS)*

The PSS (Cohen et al., 1983) consists of 14 items designed to measure the degree to which stress is experienced in different situations. The PSS measures both the psychological and physiological symptoms that arise from stress. The measuring instrument is a self-assessment questionnaire with a 5-grade Likert scale. The minimum value is 0 and the maximum value is 56, and high values correspond to high degrees of experienced stress. The PSS has good reliability (Cronbach's α 0.80). It has been translated into Swedish and been validated in Sweden. The measuring instrument also shows good psychometric data with Cronbach's α at 0.82 and split-half reliability at 0.84 (Eskin & Parr, 1996).

*The Shirom-Melamed Burnout Questionnaire (SMBQ)*

The SMBQ is often used to assess exhaustion in patients who meet the criteria for clinical burnout/ED (Melamed et al., 1992; Melamed et al., 1999; Shirom, Westman, Shamai, & Carel, 1997). A Swedish study concluded that the instrument meets the requirements of modern test theory and can thus be used to identify potential cases of clinical burnout/ED (Lundgren-Nilsson, Jonsdottir, Pallant, & Ahlborg, 2012). The SMBQ is a self-assessment instrument that meets the requirement of being general (non-occupational specific) and has been used both in Sweden and in other countries. (Grossi, Perski, Evengard, Blomkvist, & Orth-Gomer, 2003; Melamed et al., 1999). The instrument consists of 22 items forming four subscales with the factors Emotional exhaustion and physical fatigue, Listlessness, Tension, and Cognitive weariness. The items are rated on a seven-grade scale. Each individual gets a total result for burnout (average value of all the questions answered) and a result

(average value) for each subscale. Higher values correspond to more burnout symptoms. The SMBQ-Global is 3.75, where high burnout, pathologic, is ≥ 4.47 and low burnout, healthy, is ≤ 2.75. Good psychometric data with Cronbach's alpha values of 0.95–0.98 for SMBQ-total, and for the subscales Emotional exhaustion/fatigue 0.90–0.93; Listlessness 0.84–0.89; Tension 0.78–0.89; and Cognitive difficulties 0.94–0.97, have been reported (Lindahl Norberg, 2007).

*Quality of life (QoL) and health-related quality of life (HRQoL)*

The concept of QoL lacks one single definition, despite many attempts. The World Health Organisation's (WHO) definition of QoL seems to be the most authoritative and comprehensive one, and is currently used in most scientific contexts. The WHO divides QoL into the following six different domains: physical health, psychological health, level of independence, social relationship, environment, and personal values and beliefs (World Health Organization, 1994).

The concept of HRQoL is a narrower term encompassing the first four domains (physical health, psychological health, level of independence, social relationship) of the broader term QoL. Moreover, HRQoL encompasses aspects directly related to a person's health and can therefore be affected by interventions and medicines. Hence, HRQoL measurements intend to measure the abovementioned domains through self-assessment questionnaires. Today, the term HRQoL is often used synonymously with the general term QoL. HRQoL assessments can be used as a common measuring tool for understanding how different health interventions and medicines affect perceived health. Moreover, patient groups can use these results to form their own opinion of which interventions are valuable (World Health Organization, 1994).

HRQoL was measured with the Short-form 36 (SF-36), which is a validated, well-known and widely used generic HRQoL questionnaire with good reliability (Dempster & Donnelly, 2000; Persson, Karlsson, Bengtsson, Steen, & Sullivan, 1998; Ware & Gandek, 1998). The parents filled out a questionnaire at the start and end of the intervention. It consists of 36 questions measuring eight dimensions of HRQoL: four highlight physical function and four mental function. The eight dimensions are Physical functioning (PF), Role functioning – physical causes (RP), Bodily pain (BP), General health (GH), Vitality (VT), Social functioning (SF), Role functioning – emotional causes (RE) and Mental health (MH). Furthermore, Physical Component Summary (PCS) and Mental Component Summary (MCS) scores were calculated. According to Sullivan et al.

(Sullivan, Karlsson, & Taft, 2002) 80–85% of the total variance in the eight subscales can be explained by the two summary scores. All SF-36 measurements are norm-based, with the general population mean equal to 50 and the standard deviation equal to 10.

A questionnaire was created where clients answered questions about their life satisfaction. The questionnaire had questions about the clients' spare time, relation to child, relation to partner, relation to friends, and satisfaction with work. Respondents rated their experiences on visual analogue scales of 0–10 centimetres where 0 equals "not satisfied at all" and 10 equals "very satisfied".

### Treatment, Studies II and III

The two groups were offered either a structured CBT intervention or a structured mindfulness intervention. The CBT intervention for Stress and Burnout was based on standard CBT, SIT and stress management techniques (Meichenbaum & Novaco, 1985; O'Donohue & Fisher, 2008) as well as upon Acceptance and Commitment Therapy (ACT) techniques (Hayes, 2004) in which mindfulness and acceptance techniques were excluded. The structured mindfulness programme Here and Now Version 2.0 was developed by Dr. Ola Schenström (Schenström, 2011) and based on MBSR (Kabat-Zinn, 1990).

Both group treatments went on for eight weeks, with one two-hour session per week: all participants also committed to practise at home for 20 minutes per day with the help of self-instructing material for the duration of the intervention. The participants of the mindfulness group received a CD with guided assignments at the start of therapy. The participants of the CBT group received home assignments related to each session. The two therapists conducting the respective intervention had undergone basic therapy training in CBT (step 1) at Karlstad University. The therapist who conducted the mindfulness intervention was also a step one mindfulness instructor.

The main purpose of the CBT intervention for Stress and Burnout was to decrease symptoms of stress and burnout by increasing participant knowledge of these conditions and bolster their coping repertoire and problem solving skills: Both their intrapersonal and individual skills, and their confidence in being able to apply their coping skills in a flexible way that meets the appraised demands of the stressful situation. The underlying concept of the intervention includes a biopsychosocial model of stress, a transactional view of stress and a strengths-based approach. The transactional perspective points out that the ability to cope with stress requires the need to alter, avoid or minimise the effect of stressors by better managing the stress-engendering environment. The

strengths-based approach focuses on helping individuals transform their distress and suffering into a more valued direction (Appendix 1).

The main purpose of the Here and Now 2.0 intervention was to decrease symptoms of stress and burnout by increasing the participants' awareness of their own experiences and what was going on in their environment. The participants learned to handle negative thoughts and feelings (emotion-focused coping, antecedent emotion regulation) through a variety of exercises aimed at increasing their ability to react with a choice and with flexibility – thus more easily finding solutions to problems (problem-focused coping). The main theme of each session is listed in Appendix 2.

### *Statistical analyses*

*Study I*

We compared the two parent groups (CNS, ALL) regarding relevant background variables using the *t*-test for independent groups for time elapsed since diagnosis and age of the child at diagnosis, the $\chi 2$ test for parental and child gender, and the Mann-Whitney U test for treatment situation. Main outcome comparisons between parents of CNS tumour patients and ALL patients were done by calculating median values item-wise; the Mann–Whitney U test was used for item-wise comparisons. To examine whether background variables had any influence on parental fears, we performed univariate one-way analyses of variance and included time elapsed since diagnosis, age of the child at diagnosis, and treatment situation as covariates in separate item-wise analyses. We used SPSS 15.0 for Windows (SPSS, Inc, Chicago, Illinois) for all descriptive and inferential analyses.

Within the CNS parent group, the outcome was presented as proportions of parents reporting high-level fear (*fairly/extremely*) for each type of fear. Fears were described by presenting frequencies and summaries for each type of fear. We analysed the impact of the child's treatment situation on parental fears at the time of assessment using the Mann-Whitney U test and compared parents of children in treatment with parents of children who had completed cancer therapy. A similar analysis was carried out for the effect of treatment situation within the ALL parent group.

*Study II*

We examined within-group changes during the baseline period and changes during the intervention periods separately. First, we investigated if the changes from pre to post during the waiting period (baseline) were statistically significant. Second, we examined separately within the CBT and Mindfulness groups, whether changes from pre to post during the intervention period were significant. All analyses were calculated using the Mplus (version 7) statistical programme (Muthén & Muthén, 1998-2015), which allows all clients taking part in the pre-measurement to be included in future analyses. The Full Information Maximum Likelihood estimation method was used on the assumption that there would be few values 'missing at random'. The pre–post changes were tested using the Wald test. Mean values and standard deviations (*SD*) were calculated using Mplus to correct the means for missing values. Thus, the corrected mean values and standard deviation are reported. The magnitude of change was reported using within-group effect size values. Due to the small number of participants, the within-group effect sizes were calculated using Hedges' *g*. The effect sizes were calculated by dividing the mean change from pre to post with the combined (pooled) *SD* (Feske & Chambless, 1995; Morris & DeShon, 2002). To avoid overestimating the treatment effects, a within-group effect size of 0.5 was considered small, 0.8 medium, and 1.1 large (Aitken et al., 2009; Roth & Fonagy, 2005).

*Study III*

The Wilcoxon signed-rank test was used for statistical analyses in Study III. It is a nonparametric test. As the Wilcoxon signed-rank test does not assume normality in the data, it can be used when the assumption has been violated and the use of the dependent *t*-test is inappropriate. It is used to compare two sets of scores that come from the same participants. For example, when we wish to investigate any change in scores from one time point to another, or when individuals are subjected to more than one condition (Saracci, 1969). When testing for clinical significance by comparing the samples with mean values of a norm population, one-sample *t*-tests was used.

## RESULTS

### Study I

#### *Background variables*

Parent groups (CNS, ALL) differed regarding treatment situation, time elapsed from diagnosis to assessment, and age of the child at diagnosis, but were similar regarding other studied background variables.

#### *Strength of different parental fears*

The outcome of the categorization that delineated low-level and high-level fear was that the most common fears for both groups (CNS and ALL) were those of relapse, late effects of treatment and physical growth of the child. These fears were experienced fairly or extremely much by approximately 40% to 90% of parents of CNS-tumour patients (Table 2, Study I).

#### *Comparisons between CNS tumour group and reference group*

The overall pattern of severity of fears was similar for parents of CNS tumour patients and reference parents of ALL patients. However, the fears of parents of CNS tumour patients generally tended to exceed those of the reference parents (Figure 1 and Table 2, Study I). Thus, parents of CNS tumour patients reported significantly stronger fear for 7 of the 11 items than reference parents of children with ALL, based on analyses unadjusted for background variables, whereas reference parents reported significantly greater fear about the child's fertility later in life than parents of the CNS group. When controlling for the background variables treatment situation, time elapsed from diagnosis to assessment and age of the child at diagnosis in univariate analyses, group differences remained. However, for the fear about the child's chances to find a partner in life, differences did not remain when age at diagnosis and treatment situation were controlled for. Examination of effects of parent gender showed that mothers' and fathers' strength of fears were approximately similar and displayed a similar pattern (Table 2, Study I).

#### *Treatment situation*

In the CNS tumour group, child still in treatment was related to greater parental fear concerning late effects of treatment ($p$ = .006), physical growth ($p$ = .002), fertility ($p$ = .017) and sexual development ($p$ = .014). Parents in the reference

group with children in treatment reported greater fear about cancer recurrence ($p = .049$).

**Study II**

***Comparative analyses of participants' symptom rates during baseline and before and after interventions.***

Table 2 in Study II shows that during the baseline period, stress as measured by PSS decreased significantly from pre to post. However, during baseline, the pre to post within-group effect size was small ($d = 0.45$) and non-significant, 95% Confidence Interval (CI) = -0.07–0.96.

Significant decreases in PSS scores were also observed for the CBT and Mindfulness groups from pre to post when the participants were offered interventions after baseline (Table 2, Study II). The within-group effect sizes were large and significant for both the CBT group ($d = 1.64$; 95% CI = 0.63–2.66) and the Mindfulness group ($d = 2.20$; 95% CI = 1.03–3.37) from pre to post. Thus, although there were positive changes in both groups regarding stress, the within group effect sizes were considerably larger in the CBT and Mindfulness groups as compared to the baseline condition.

Burnout symptoms as measured by SMBQ-Global decreased significantly both in the CBT and Mindfulness conditions from pre to post, but not during baseline (Table 2, Study II). The within-group effect seizes were large and significant for both interventions groups (CBT, $d = 1.28$, 95% CI = 0.32–2.24; Mindfulness, $d = 1.25$, 95% CI = 0.24–2.26). The within group effect size was very small and non-significant from pre to post during the baseline condition ($d = 0.03$, 95% CI = -0.46–0.54).

In summary, the results show that both the CBT and Mindfulness interventions are effective with statistically significant improvements on outcome measures with large within-group effect sizes suggesting that the changes are clinically relevant. In both groups, parents' stress and burnout symptoms were reduced. In contrast, markedly smaller changes were observed during the baseline period.

**Study III**

*Comparative analysis of SF-36 component summary scores before and after therapy*

Participant results of the SF-36 MCS scores before and after therapy indicate that there had been considerable improvement (Wilcoxon signed-rank test) from the beginning of the therapy to its conclusion for both treatments (CBT: $z$ = -2.55, $p$ = .01; Mindfulness: $z$ = -2.31, $p$ = .02). However, the results of the SF-36 PCS scores before and after therapy showed no significant improvement, that is, the effect of treatment was the same irrespective of the form of treatment received. When testing for clinical significance (Table 1, Study III) by comparing the samples with mean values from a norm population (one-sample $t$-tests), MCS scores were significantly lower at pre-measurements for the CBT and Mindfulness groups, but no significant differences were observed for post-measurements. For the PCS post-measurement of Mindfulness, a significantly higher score was observed compared to the norm population (*c.f.* Table 1, Study III). The results of the SF-36 MCS scores are illustrated graphically in Figure 1, Study III.

*Comparative analysis of SF-36 mental scales before and after therapy*

Participants' results on the SF-36 mental scales showed substantial improvements from before therapy to after therapy, meaning that the treatment effect was similar for CBT and Mindfulness in this respect. Wilcoxon signed-rank tests showed significant effects from pre- to post-treatment for the following dimensions and groups: Vitality (CBT: $z$ = -2.69, $p$ = .007; Mindfulness: $z$ = -2.32, $p$ = .02), Social functioning (CBT: $z$ = -2.53, $p$ = .01; Mindfulness: $z$ = -2.22, $p$ = .03), Role emotional (Mindfulness: $z$= -2.02, $p$ = .04) and Mental health (CBT: $z$ = -2.40, $p$ = .02; Mindfulness: $z$ = -2.40, $p$ = .02). A tendency to significant effect was also seen for the CBT group in the dimension Role emotional ($z$ = -1.86, $p$ = .06). The results of the SF-36 mental scales are illustrated graphically in Figure 2, Study III.

When testing for clinical significance by comparing the samples with mean values from a norm population, significantly lower pre-measurements were observed in both groups for the dimensions Vitality, Social functioning, Role emotional and Mental health, whereas at post-measurement only the dimension Mental health was lower for both groups compared to norm values (*c.f.* Table 1, Study III).

### *Comparative analysis of SF-36 physical scales before and after therapy*

Participants' results on the SF-36 physical scales before and after treatment were generally similar (Wilcoxon signed-rank test). The only exception was the Mindfulness group, which had improved from pre- to post-treatment in the dimensions Role physical ($z$ = -1.93, $p$ = .05), Bodily pain (z = -2.04, $p$ = .04) and General health ($z$ = -2.32, $P$ = .02). A tendency to significant effect was also seen for the CBT group in the dimensions Role physical ($z$ = -1.79, $p$ = .07) and General health ($z$ = -1.79, $p$ = .07).

When testing for clinical significance by comparing both groups with a norm population, it could be concluded that our sample did not differ from the norm population on the physical dimensions of SF-36, except in the General health dimension where both groups scored lower at pre-measurement but scored normally at post-measurement (*c.f.* Table 1, Study III). The results of the SF-36 physical scales are illustrated graphically in Figure 3, Study III.

### *Comparative analysis of life satisfaction before and after therapy*

For life satisfaction, considerable improvements (Wilcoxon signed-rank test) had taken place during the therapy for the scales Spare time (CBT: $z$ = -1.99, $p$ = .05), Relation to child (CBT: $z$ = -2.40, $p$ = .02; Mindfulness: $z$ = -2.20, $p$ = .03) and Relation to partner (CBT: $z$ = -2.52, $p$ = .01). The results for life satisfaction are illustrated graphically in Figure 4, Study III.

## GENERAL DISCUSSION

The general aim of the present research was to survey parental fears and investigate HRQoL, stress and burnout in parents of children with chronic conditions treated with mindfulness and CBT. Parental fears were generally high in all investigated families, but higher in families of children who had survived brain tumour, compared with a reference group of parents of leukaemia survivors. CBT and Mindfulness were effective with statistically significant improvements on nearly all outcome measures. For instance, the parents' stress and burnout were reduced in both groups. The results in Study III indicate significant improvements for parents in the two treatment groups regarding MCS scores, Vitality, Social functioning and Mental health. Considerable improvements have also taken place for Life Satisfaction and Relation to child.

### Parental fears

Study I portrays how illness-related fears are experienced by parents of children diagnosed with a brain tumour. Fear concerning a future cancer recurrence and fear concerning late effects of treatment were most prominent among all parents, but more so among parents of CNS tumour patients.

The strongest fear of parents of all children was that of a future relapse, and even higher in parents of children treated for a CNS tumour. This reflects the fact that, despite the success of modern therapies, long-term outcome remains a source of parental uncertainty. This finding supports recent research addressing FOR (Fidika et al., 2015). Two aspects of uncertainty are important to highlight: hoping and planning. Despite being hopeful of about the future, these parents live in a world of uncertainty and are afraid to dream about the future (Cantrell & Conte, 2009).

The continuous need for follow-ups to check for any recurrence of the tumour serves as a reminder to parents about the unpredictability of the final outcome (Eiser, Richard Eiser, & Greco, 2002). Hence, a justified coping strategy for parents is the 'wait and see' approach (Benesch et al., 2006). Here, the actual risk may play a role because long-term survivors of childhood CNS tumours are 13 times more likely to die prematurely than healthy age–sex matched peers, and illness recurrence remains the most common cause (70%) of their deaths (Sklar, 2002). Moreover, parental fears about their child's school performance could reflect that cognitive impairments, caused by the illness and its treatment, are common among children treated for CNS tumours – and cognitive

dysfunction could be more limiting than physical effects in the integration of cancer survivors into mainstream society (Aarsen et al., 2006; Fouladi et al., 2005; Mulhern et al., 2005).

The fear of a complete decline in health is an indistinct question, although with an unquestionable reference to a most threatening negative outcome of some kind. The fear of a complete decline could be interpreted as fear of a severe chronic condition or as fear of the death of the child. Maybe this fear could be seen as a shift in existential outlook. It relates to an understanding that even the most unlikely and the most unwanted can happen, which is a reasonable trigger of fear and distress. Also, fear of a complete decline reflects that the long-term outcome remains a source of parental uncertainty. A number of studies show that parents of children treated for cancer report higher levels of distress (Boman et al., 2003; Norberg & Boman, 2008; Sultan et al., 2016), with parents of children of CNS tumours being a particularly vulnerable group for heightened distress (Hovén et al., 2008). A substantial number of parents in the present study reported great fear of health-related late effects of treatment. Such concerns appear to be adequate as many adult survivors of childhood CNS tumours suffer from persistent deficits in cognition, sensory function, mobility and self-care (Boman, Hovén, Anclair, Lannering, & Gustafsson, 2009).

Findings from Study I indicate that parental fear of treatment-related sequelae is greater when a child is still receiving active cancer treatment. We can assume that parents of a child for whom treatment is already completed have reached a point where the initial shock has passed. There are indications that parents of children who have been off treatment for a considerable length of time experience uncertainty about the final severity of actual sequelae, as well as about possible new upcoming problems (Hovén et al., 2008; Norberg, 2010). After cessation of treatment, parents have to adapt to living with uncertainty about the recurrence of illness as well as possible long-term late effects. Such uncertainty about the final outcome, together with new stressors, could lead to the feeling of having lost control of events, to passivity in the face of stress and to dysfunctional coping with stressors – which may result in chronic stress reactions (Lindahl Norberg, 2007; Lindström et al., 2010; Norberg, 2010).

**CBT and Mindfulness effects on stress and burnout**

The result of Study II shows that perceived stress, and the degree to which situations are perceived as stressful, and burnout, such as emotional exhaustion and physical fatigue, listlessness, tension and cognitive weariness, all decreased

significantly irrespective of the form of treatment received (CBT or Mindfulness).

It was interesting that both interventions seemed to work equally well on parents of children with chronic conditions. There may be several possible explanations for these results. One, previous research shows that both mindfulness and CBT are effective interventions for reducing stress-related illness in general (Goyal et al., 2014; Lewis & Lewis, 2016). Two, we cannot exclude that other interventions would have worked because the study lacks an independent control group. There is also the possibility, even if it is small considering the results of the waiting list control, of a spontaneous recovery effect.

Our CBT intervention appeared to have an effect on stress and burnout, which can be explained on the basis of previous research on CBI, SIT and stress management (Hofmann et al., 2012; Meichenbaum & Novaco, 1985; O'Donohue & Fisher, 2008) and ACT (Hayes, 2004). As the Cochrane review (Eccleston et al., 2012) suggested, our intervention focused on bolstering clients' coping repertoires and problem solving skills through different well- established CBI and SIT stress management techniques such as cognitive reframing, perspective taking problem-solving, relaxation, emotion regulation (Beck, 1964; Ellis, 1973) and valued direction (Hayes, 2004). Another underlying concept of the effectiveness of our CBT intervention can possibly be explained by the fact that the aforementioned techniques increase psychological flexibility. For example, being flexible and willing to engage in difficult activities in order to continue in the direction of gaining important insights allows a person to pursue a rich, meaningful life right away, which in turn leads to less distress and increased psychological flexibility (Hayes, 2004).

One explanation why mindfulness seems to have an effect on stress and burnout for these parents can be explained by the underlying mechanisms of mindfulness. The participants learned how to handle negative thoughts and feelings (emotion regulation, emotion-focused coping) through exercises that increased their ability to react with a choice and with flexibility and thus more easily find solutions to problems (problem-focused coping). Broadly, the participants' goal was to learn to observe a greater variety of experiences and sensations (non-avoidance coping) and describe their thoughts and feelings without judging or suppressing (response-focused emotion regulation). Developing mindfulness skills leads to non-judgemental and non-reactive acceptance of all experience, which in turn results in positive psychological

outcome (Kabat-Zinn, 1982; Kabat-Zinn et al., 1992; Segal, Williams, & Teasdale, 2002). Several studies have shown that mindfulness reduces rumination and worry and increases psychological flexibility (Cahn & Polich, 2006; Siegel, 2007a). And psychological flexibility is, in turn, seen as a fundamental aspect of health and well-being (Kashdan & Rottenberg, 2010). Meditation also activates the brain region associated with more adaptive responses to stressful or negative situations (Cahn & Polich, 2006; Davidson et al., 2003). Activation of this region corresponds with faster recovery to baseline after being negatively provoked (Davidson, 2000; Davidson, Jackson, & Kalin, 2000). Other important aspects are that mindfulness meditation has a positive effect on relationship satisfaction and protects against the emotionally stressful effects of relationship conflict (Barnes, Brown, Krusemark, Campbell, & Rogge, 2007).

Mindfulness may also contribute to effective emotion regulation strategies (Doll et al., 2016; Opialla et al., 2015). Further, people who observe their thoughts and feelings with openness and curiosity show a different activation pattern, with labelling linked to greater prefrontal cortex activity and a simultaneous inhibition of the limbic responses, which in turn are bound to executive functioning (DeYoung, Peterson, & Higgins, 2005; Kalisch et al., 2005).

Paul Gilbert has integrated biological, evolutionary and social psychology theories of how people regulate emotions into his compassion-focused therapy (Gilbert, 2014a). Pedagogically, he explains how complex psychological and biological systems interact and can be explained from three different systems: the alarm system, the drive system and the soothing system.

The fight, flight and freeze responses of the alarm system are activated in response to a perceived threat, and their function is to get us to find protection (Frijda, 1988; Selye, 1991). Feelings linked to this system are worry, anxiety, stress, fear, anger and shame (Gilbert, 2014b). The drive system is an activating and motivational system that strives to achieve rewards and activates wellness. It gives rise to joy, excitement and pleasure. The soothing system is activated when the other two systems are passive, that is, when we do not strive to achieve something or need to deal with various kinds of threats. When this system is activated, we experience feelings of peace, joy and satisfaction (Gilbert, 2014a, 2014b).

People, particularly those with a high degree of self-criticism and shame, can have an overactive alarm system (Gilbert, 2014a). The soothing system is then often inactive and difficult to access. In these cases, the systems no longer fulfil their functions – to protect us from threats and act purposefully. The

consequences of an overactive alarm system are lack of recovery, difficulty regulating emotions, passivity and avoidance coping, which in worst case lead to long-term stress and fatigue. However, the soothing system can be reactivated through, for example, mindfulness and CBT techniques, which balance up negative feelings and thoughts and calm down the alarm system (Gilbert, 2014b). At best, it results in functional coping, improved emotion regulation strategies and increased psychological flexibility. This hopefully means the parents can handle all the demands and stressors that come with being a parent of a child with a chronic condition.

## HRQoL

The feasibility of CBT and Mindfulness interventions for parents of children with chronic conditions were studied in Study III, with a focus on HRQoL and life satisfaction. The results indicate significant improvements for parents in the two treatment groups regarding MCS scores, Vitality, Social functioning and Mental health. These findings demonstrate enhanced HRQoL in terms of parents' functionality and well-being. No significant effects were found for the other variables relating to physical health (PCS scores and Physical functioning). The fact that no significant effects were found for the physical variables is in agreement with earlier investigations stating that the effect of CBT on physical health is uncertain (Roy-Byrne et al., 2010), which agrees with another study of ours where the focus was on problem areas such as depression, anxiety, self-esteem and multiple problems (Henriksson, Anclair, & Hiltunen, 2016). There was also convincing consistency between the study by Henriksson et al. (2016) and the study by Rufer et al. (2010) on most physical subscales, where CBT seems to have limited effect. The same conclusion can be drawn from a comparison with Niles et al. (2013) when evaluating the index value of physical health. Our population did not differ from the norm population on the physical dimensions on SF-36, except for GH, which means that the possibility of improvement was significantly less for physical dimensions. For the mindfulness group, improvements could also be seen for Role emotional as well as for the physical dimensions Role physical, Bodily pain and General health. These results indicate that mindfulness can reduce physical symptoms of stress, thereby giving other effects on HRQoL than CBT for parents of children with chronic conditions. Moreover, improvements in life satisfaction regarding Spare time and Relation to partner were seen for the CBT group, and both groups improved their life satisfaction with regard to Relation to child.

In addition, the results of the present study show low satisfaction of life initially and improvement over time regarding parents' Spare time, Relation to child and Relation to partner. These results are partly in agreement with the findings of Lawoko and Soares (2002), which showed that parents of children with congenital heart disease or other diseases have a lower QoL compared with parents of healthy children. These findings support the hypothesis that parents of children with chronic conditions suffering from stress and burnout can enhance their daily functioning and well-being and alleviate psychological symptoms of these conditions by participating in either an 8-week CBT intervention for Stress and Burnout or in a mindfulness programme based on MBSR called Here & Now 2.0.

**Gender**

The majority of patents included in the abovementioned studies were mothers. There may be many reasons why mothers, to a greater extent than fathers, seek out treatments for stress and burnout. Several studies show that mothers report lower QoL than fathers and that reduced QoL depends on care-giving time, sick-leave, retirement, financial difficulties, distress, hopelessness and social isolation (Hovén, von Essen, & Norberg, 2013; Lawoko & Soares, 2002, 2003). One explanation is that mothers do not feel that they have the time or the opportunity to focus on themselves and their health, which among other things have led to their current state of burnout – with sick leave as a result (Anclair & Hiltunen, 2014; Hovén et al., 2017). Another study shows that mothers' well-being is dependent on their feeling unconditionally loved, feeling comforted when in distress, authenticity in relationships and satisfaction with friendships (Luthar & Ciciolla, 2015). These important life areas will likely suffer when mothers have little time to prioritise themselves and are on sick leave. A recently published licentiate thesis investigated parental stress in relation to parental leave. The participants consisted of 280 parents who answered the Swedish Parenting Stress Questionnaire on perceived parental stress 6 months and 18 months after the birth of their child. The results showed that parents who did not share parental leave equally reported higher parental stress 18 months after child birth, compared to parents with equally shared parental leave (Lidbäck, 2016). Hence, in cases where the child also suffers from a chronic disease, it may therefore be of particular importance for healthcare practitioners to emphasise the importance of shared parental responsibility to prevent stress and burnout in mothers. In the present study, a possible reason for the parents' improvement both in mental health and in general health may be that, as a result of the

intervention, they can access their strengths and act functionally and flexibly to better manage the demands and additional stressors that come with being a parent of a child with a chronic condition.

**Clinical implications**

Clinical experience shows that this parent group (mostly mothers) visit healthcare centres due to pain in the head, shoulders and/or neck; anxiety; depression and sleep problems, which become the focus of treatment (e.g. medical treatment or physical therapy). However, as a consequence of having a child with a chronic illness, many of the parents meet the criteria for burnout or ED – which is rarely recognised or treated. One study concludes that 98% of people with burnout/ED suffer from at least one somatic symptom, the most common symptoms being nausea, gastrointestinal problems and headaches (Glise et al., 2014).

In addition, many parents wait too long before seeking treatment for their symptoms. One study investigating the course of burnout/ED shows that a long duration of symptoms before consultation with healthcare was associated with a prolonged time of recovery and that the recovery period of different patient groups with stress-related mental health problems can differ considerably (Glise, Ahlborg, & Jonsdottir, 2012). One plausible explanation could be ongoing stress exposure or new stressors during the course of illness and rehabilitation (Glise et al., 2012). This is often the case in parents of children with chronic conditions: new stressors arise due to the child's illness and treatment. To be able to work preventively with psychoeducation about stress and burnout for parents of children with chronic illness, it is of utmost importance that healthcare services have better knowledge of how common ED is in this parent group. It is crucial to identify these parents when they seek healthcare for somatic symptoms, anxiety and depression and offer interventions that not only focus on the specific symptom, but psychological interventions that focus on factors that may lead to stress and burnout. Examples of such factors are fear/anxiety; catastrophising; increased sympathetic activation; emotional avoidance; lack of recovery; lack of social support; poor problem-solving skills, coping strategies and emotion regulation; and lack of psychological flexibility.

## Methodological considerations

### *Representativity and generalizability*

The large number of parents included in Study I ensures that the findings obtained are representative of the population of parents from which the sample was taken. Hence, the inclusion procedures of Study I were sufficiently rigorous to ensure an unbiased selection of parents.

Studies II–III have certain limitations concerning representativity. Parents were recruited through advertisement in the local newspaper in Karlstad. Advertising for participants may, however, lead to a non-representative sample: The participants recruited could be more or less ill than others, or have a different background or lifestyle that could affect the outcome.

Due to the restricted population size in Study II, this pilot study had to settle for 19 participants when investigating the effects of the interventions. Although significant results were observed, the reader needs to observe that several parents dropped out after the baseline period. Low-powered studies may also result in an overestimation of effect sizes or in non-replicable effects. Further, the parents constituted their own controls and were wait-listed for six months, which were not ideal control conditions.

Finally, another question of representativity concerns non-responders and dropouts in the studies. Data on the non-responders in Study I and dropouts in Studies II–III were not available.

The dropout rate in Study II was 32,1% after the baseline period, before the interventions were offered. This is consistent with research literature on dropout analysis (McFarland & Klein, 2005). The bulk of psychotherapy research is pursued in atypical settings such as academic centres and student clinics. Data from such settings show that 25–50% of the participants drop out of psychotherapy interventions during the course of the first five sessions. Research available in the field shows that early dropout is associated with lower age, low income, low education, substance abuse and insufficient social support (Centorrino et al., 2001; Foulks, Persons, & Merkel, 1986; McFarland & Klein, 2005; Pekarik, 1992; Renk, Dinger, & Bjugstad, 2000; Sparr, Moffitt, & Ward, 1993). A study by Crane and Williams (2010) indicates that persons with high levels of cognitive reactivity, depressive rumination and brooding have difficulties completing and engaging in MBCT. Paradoxically, these are the persons who may benefit the most from completing MBCT. It is therefore of utmost importance to figure out how to motivate and support this group to complete MBCT treatment. A review by Santana et al. (2011) highlights that

between 10% and 57% of the anxiety patients drop out before the end of treatment. Comorbidity with depression was the factor that caused the greatest dropout rate. For parents of children with chronic illness, the most common reason reported for not participating has previously been lack of time (Landolt, Boehler, Schwager, Schallberger, & Nuessli, 1998).

### *Assessment validity and reliability*

A theoretical, problematic issue in Study I was comparing different childhood malignancies in terms of their impact on parents' illness-specific fears. Both groups experienced fears, and the diagnostic groups differed greatly. Despite the fact that the fears studied were found to be the most characteristic in both populations, other fears not covered by our questionnaires may be of particular significance for the individual parent.

In Studies II and III, conclusions about treatment effects cannot be ensured due to the uncontrolled design. The within-group effect should therefore be interpreted with caution as it is possible that the improvement in the two groups was caused by spontaneous recovery. However, results from earlier studies suggest that such a large spontaneous recovery is unlikely (Brinkborg, Michanek, Hesser, & Berglund, 2011). Another important aspect that may have affected the internal validity is placebo effects. The participants were randomly selected to one of the interventions and did not know in which intervention they took part. Another possible limitation of the studies concern the self-report evaluation. Different response styles may be systematically prevalent in different groups. A response-shift bias may thus have influenced the ratings.

### In summary

Worries and uncertainty about the progression or outcome of the child's condition, together with new stressors, may result in feelings of having lost control of events, passivity in the face of stress, dysfunctional coping with stressors and difficulties with emotion-regulation. This in turn may lead to chronic stress reactions, especially in mothers of children with chronic conditions (Vrijmoet-Wiersma et al., 2008). Both our CBT intervention for Stress and Burnout and the mindfulness programme Here & Now 2.0 appear to have an effect on stress and burnout. One explanation why both interventions seem to have an effect on stress and burnout for these parents can be explained by theories of stress, emotions and coping (Cohen & Wills, 1985; Frijda, 1988; Gross, 2002; Karasek & Theorell, 1990; Lazarus & Folkman, 1984; Selye, 1950). Both interventions, but from different perspectives, focus on reducing

symptoms of stress and burnout by bolstering participants' coping and problem solving skills, emotion regulation strategies and ability to apply their skills in a flexible fashion that meets the appraised demands to stressful situations, as suggested by the Cochrane review (Eccleston et al., 2012). Another underlying reason for the effectiveness of both our CBT intervention and the Here & Now 2.0 mindfulness programme can possibly be explained by the fact that these techniques increase psychological flexibility. Psychological flexibility is seen as a fundamental aspect of health and well-being (Kashdan & Rottenberg, 2010). To be able to work preventively with psychoeducation about stress and burnout for parents of children with chronic illness, it is of utmost importance that healthcare services have better knowledge of how common ED is in this parent group. It is crucial to identify these parents when they seek healthcare for somatic symptoms, anxiety and depression and offer interventions that not only focus on the specific symptom, but psychological interventions that focus on factors that may lead to stress and burnout. Examples of such factors are fear/anxiety; catastrophising; increased sympathetic activation; emotional avoidance; lack of recovery; lack of social support; poor problem-solving skills, coping strategies and emotion regulation; and lack of psychological flexibility. Findings from this thesis support the hypothesis that parents of children with chronic conditions suffering from stress and burnout can enhance their HRQoL, daily functioning and well-being, as well as alleviate psychological symptoms of stress and burnout by participating in an 8-week CBT intervention for Stress and Burnout or in a mindfulness programme based on MBSR called Here & Now 2.0.

## CONCLUSIONS

Fears are common among parents of children with cancer, and the most prominent fear is the recurrence of illness and late effects of illness and treatment.

Group treatment with either CBT for Stress and Burnout or the mindfulness programme Here & Now 2.0 can be efficient for reducing stress and burnout in parents of children with chronic conditions.

Group interventions based on CBT or mindfulness improve parents' perceived HRQoL.

Satisfaction of life improved over time regarding parents' Spare time, Relation to child and Relation to partner.

It is important to find out if there is a child with a chronic illness in the family when a person seeks treatment for stress-related symptoms in order to detect and diagnose burnout in time.

Efforts should be targeted to burnout/ED rather than to specific symptoms, such as pain and anxiety, to prevent these patients from entering into the acute phase of burnout – resulting in a long recovery period and sick leave.

It is essential to inform parents of children with chronic illness that they are at higher risk of developing burnout syndrome than parents who do not have a child with a chronic disease.

It is our hope that our CBT for Stress and Burnout intervention and our Here & Now 2.0 mindfulness programme will be used more extensively (e.g. in child healthcare and mental health nursing) and that they will become generally available to parents suffering from parental stress and burnout.

In a wider perspective, children will benefit from having parents who have learnt to identify stressors and found functional coping and emotion regulation strategies preventing the stress from turning into clinical burnout or depression.

## FUTURE PERSPECTIVES

Directions for forthcoming studies should include RCTs comparing MBI, CBT and treatment as usual, as well as longitudinal follow-up to see if the effects last over time. Future research should also offer interventions that can be implemented via the Internet to reach parents who find it difficult to set off time for treatment.

 The psychological and emotional consequences of being parents of children with chronic disease can be further investigated through more complex assessment tools examining, for example, sleep patterns, coping strategies, emotion regulation strategies, psychological flexibility and recuperation, but also comorbidity with other mental illnesses.

Preventive interventions for parents of children with chronic illness should focus on recuperation, physical activity, flexible coping skills and adaptive emotion regulation strategies, as well as on shared parental leave, to prevent parents from suffering burnout and chronic stress.

It is also important to investigate the relationship between parents' and their children's HRQoL to see if the children benefit from interventions targeted for parents. Moreover, it is crucial to find out which components, in different interventions, affect outcome to be able to tailor interventions for parents of children with chronic illness suffering from stress and burnout. Of particular interest in future research is to investigate whether psychological flexibility can be seen as a mediator of reduced stress and burnout and, if that is the case, specifically design interventions with such a focus.

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## Appendix 1.

**CBT for Stress and Burnout**

The CBT intervention includes a conceptual educational phase, weeks 1–2; a skills acquisition and consolidation phase, weeks 3–7; and an application phase, week 8. Weeks 1–2 include psychoeducation about stress and burnout according to the biopsychosocial model of stress, differences between changeable and unchangeable aspects of stressful situations, and collaboratively identified goals. During weeks 3–7, the participants practise problem-focused and emotion-focused coping skills through different CBT techniques such as cognitive reframing, perspective taking, problem-solving, relaxation, emotion regulation and behavioural practice. The last week focuses on relapse prevention and on applying all techniques in everyday life.

### *Session 1 Introduction to stress and burnout*

Brief outline of CBT as a therapy method
On stress and exhaustion
Purpose and aim of the treatment
Home assignment: Daily self-observation of stress behaviour

### *Session 2 Formulating personal goals*

Follow-up on home assignment
Overriding personal goals with the treatment
What I want to achieve and how to achieve it
Group task: Identify goals
Home assignment: Identify goals that are realistic and specific
Continued observation of stress behaviour

### *Session 3 On thoughts*

Follow-up on home assignment
Thoughts: Negative automatic thoughts
Question negative automatic thoughts
Thinking traps
Situation–thought–behaviour–consequence
Group task: How stress affects thoughts, thinking traps
Home assignment: Daily registering of situation–thought–behaviour–consequence

### Session 4 On emotions

Follow-up on home assignment
On emotions
The function of emotions
Emotion spotting
Situation–thought–emotion–behaviour–consequence
Group task: How stress affects emotions
How emotions affect me and with what consequences
Home assignment: Daily registering of situation–thought–emotion–behaviour–consequence

### Session 5 Problem solving

Follow-up on home assignment
Situation–thought–emotion–behaviour–consequence
Problem-solving techniques
Group task: Problem solving: define a problem; brainstorm for solutions; make a plan and, when completed, decide if the problem is solved or not.
Home assignment: Problem-solving training; identify situations, thoughts and feelings that you avoid; and register situation–thought–emotion–behaviour–consequence

### Session 6 Valued direction

Follow-up on home assignment
Life compass: Parenthood, work, health and social activities
How to act in a valued direction on the basis of valued direction and treatment goals.
Group task: How to act in accordance with a valued direction
What prevents me, and how can I solve the problems?
Home assignment: Make your own life compass and start acting on it

### Session 7 Activity/tension, relaxation and recovery

Follow-up on home assignment
Psychoeducation on relaxation/tension and recovery
Understanding the difference between overactivity/underactivity
Group task: Tension exercise: discover the difference between tension and relaxation
Home assignment: Daily relaxation breaks, problem-solving and acting accordingly

### *Session 8 Moving on*

Follow-up on home assignment

Everyday applications of the whole manual

Group task: Design routines to maintain the programme, identify risks and move on.

## Appendix 2.

**Here and Now 2.0 Mindfulness for Stress and Burnout**

Here and Now 2.0 includes the following basic elements: body scan, breathing anchor, being aware of breathing and body movement, mindful movements and sitting meditation. These are introduced during weeks 1–4. The techniques are trained alternately in weeks 5–8, and the Stop, Observe, Accept, Let go (SOAL) model is integrated in the training during these weeks.

### *Here and Now 2.0 Mindfulness for Stress and Burnout*

Session 1 Get to know your body – Body scan
Introduction to the Here and Now Programme
Survey of body scan
Eating a fruit and being consciously present
Home assignment: Body scan and, for example, eating a fruit daily and being consciously present

### *Session 2 Observe your breathing – Breathing anchor*

Follow-up of the week's experience of home assignments
Breathing exercise with a focus on your breathing
Exercise the breathing anchor
Survey the relationship between thought–body–feeling and mindfulness, and their interplay
Home assignment: Body scan and daily breathing anchor

### *Session 3 Being aware of breathing and body movement*

Follow-up of the week's experience of home assignments
Talk about possible resistance and obstacles for performing the exercises
Breathing exercises
Mindful yoga
Home assignment: Mindful yoga
Body scan
Breathing exercises morning and evening

### *Session 4 Just sit – Here-and-now sitting meditation*

Follow-up of the week's experience of home assignments
Brief outline of the research situation on mindfulness and the brain
Psychoeducation on stress
Practise sitting meditation
Home assignment: Mindful yoga

Sitting meditation and breathing exercises morning and evening

### Session 5 Acceptance: Stop, Observe, Accept, Let go (SOAL)

Follow-up of the week's experience of home assignments
Get familiar with SOAL
Practise sitting meditation

### Session 6 Coping with difficulties

Follow-up of the week's experience of home assignments
Get familiar with SOAL

### Session 7 Thoughts are not facts

Follow-up of the week's experience of home assignments

### Session 8 Reinforce your daily training and take care

Follow-up of the week's experience of home assignments

# Fears, Stress and Burnout in Parents of Children with Chronic Conditions

The aim of the present research was threefold: to investigate the fears of parents of children with chronic conditions; to evaluate the effectiveness of their treatment with either mindfulness-based therapy or cognitive behavioural therapy (CBT); and to assess treatment outcome in terms of health-related quality of life (HRQoL). Long-term stress can lead to some form of chronic stress reaction. In study one, fears of future cancer recurrence and of late effects of treatment were most prominent among parents of CNS tumour patients. Study two investigated the effectiveness of two group-based interventions on stress and burnout among parents of children with chronic conditions. Parents were offered either a CBT or a mindfulness programme. Both interventions significantly decreased stress and burnout. Study three focused on the HRQoL and life satisfaction of the parents in study two. The results indicate improvements for participants in both treatment groups regarding certain areas of HRQoL and life satisfaction. To conclude, fears concerning future cancer recurrence and late effects of treatment are most prominent among parents of children with cancer. Another conclusion is that CBT and mindfulness decrease stress and burnout and may have a positive effect on areas of HRQoL and life satisfaction.