Chapter 5
The Role of Perfectionism in Chronic Fatigue Syndrome

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Introduction

Nancy, a 42-year-old woman, has been suffering from disabling fatigue and general weakness for the past 5 years. Over the last 6 months, her symptoms have gradually become worse. She has had repeated viral throat infections and was exhausted almost all of the time, which forced her to give up her demanding job. She used to be very active (e.g. in sports) despite her fatigue complaints, but now she can only carry out simple daily tasks, and even small amounts of exercise (e.g. doing household work) typically lead to extreme exhaustion or a complete energy "crash". Her social life became severely restricted by the illness. After a comprehensive examination at the hospital including several laboratory tests, no medical (or psychiatric) condition could be identified that may explain her complaints. She was then diagnosed with chronic fatigue syndrome (CFS). Nancy's story is typical of many patients diagnosed with CFS.

CFS is a complex syndrome that is still the subject of much controversy which is in large part due to the fact that relatively little is known about the exact causes of this disorder. As illustrated by the story of Nancy, the syndrome is usually associated with severe functional impairment and social isolation. As a result, CFS places an enormous economic burden on health-care systems and society in general (Jason,

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Benton, Valentine, Johnson, & Torres-Harding, 2008; Reynolds, Vernon, Bouchery, & Reeves, 2004). For instance, the annual productivity loss related to CFS in the USA is estimated at approximately $ 9.1 billion (Reynolds et al., 2004).

We begin this chapter with a brief description of the diagnosis of CFS. We then present a working model for the development and maintenance of CFS integrating biological and psychological research findings. Next, we specifically focus on the purported role of perfectionism in CFS, the topic of this chapter. We provide an overview of theory and research on perfectionism in CFS. After reviewing empirical studies that have investigated the role of perfectionism in CFS, we look at the mechanisms that may explain the relationship between both. We then look at the possible origins of perfectionism in CFS, including studies that have demonstrated a link between early childhood adversity and CFS. We close this chapter by a summary of treatment implications of these findings.

Chronic Fatigue Syndrome

The term chronic fatigue syndrome was coined in the 1980s, a century after George Miller Beard introduced the concept of neurasthenia to denote a similar condition of exhaustion (Wessely, Hotopf, & Sharpe, 1998). Today, CFS is seen as belonging to a spectrum of functional somatic syndromes, an umbrella term encompassing a group of syndromes characterized by persistent somatic symptoms (e.g. fatigue, musculoskeletal pain, and gastrointestinal symptoms) without any organic impairments (Wessely et al., 1998). Studies indicate that functional somatic symptoms are highly prevalent and may represent up to 30 % of all primary care consultations (Kirmayer, Groleau, Looper, & Dao, 2004). CFS involves both physical and mental exhaustion and usually develops gradually over months or years (most often between ages 40–49), although acute onset of CFS has also been reported (Boneva et al., 2011; Van Houdenhove et al., 2009). CFS is more common in females than in males in adults as well as in children and in adolescents (Farmer, Fowler, Scourfield, & Thapar, 2004; Gallagher, Thomas, Hamilton, & White, 2004; Jason et al., 1999). The diagnosis of CFS is based on consensus criteria developed by experts and involves an extensive medical and psychiatric examination to rule out other conditions that may be causing the symptoms. The most widely used diagnostic criteria are the Centers for Disease Control and Prevention (CDC) criteria (Fukuda et al., 1994), which define CFS as a condition that is characterized by unexplained prolonged fatigue for at least 6 months that is not the result of ongoing exertion and is not substantially relieved by rest, along with other symptoms such as musculoskeletal pain, sore throat, headaches, tender lymph nodes, substantial impairment of short-term memory or concentration, unrefreshing sleep, and post-exertional malaise (i.e. extreme exhaustion or flu-like feelings after physical and/or mental activity). In the recently redefined International Consensus Criteria (Carruthers et al., 2011), post-exertional exhaustion, that is, increased physical and/or cognitive fatigability in response to exertion, plays a more central role in the conceptualization of CFS. There is indeed

increasing evidence that effort intolerance is one of the key diagnostic features of CFS, probably reflecting a loss of "adaptability" of the human stress response system involving a reduced tolerance for a wide range of physical and mental stressors and environmental stimuli (e.g. loud noise, bright lights, alcohol, and certain foods) (Van Houdenhove & Luyten, 2010; Van Houdenhove, Luyten, & Kempke, 2013), as discussed in greater detail below.

Comorbidity with other medical and psychiatric disorders is often observed in CFS patients. For instance, studies have demonstrated a high rate of comorbidity between CFS and chronic pain syndromes such as fibromyalgia syndrome (FMS), a condition that is mainly characterized by widespread muscle pain and tenderness (Ablin et al., 2012; Kanaan, Lepine, & Wessely, 2007). In the context of the ongoing debate as to whether medically unexplained syndromes should be considered as discrete diagnostic entities or as variants of one single functional somatic syndrome (known as the "lumpers" versus "splitters" debate) (Kanaan et al., 2007), we and others have argued that both conditions may comprise one larger cluster of fatigue and pain symptoms, particularly because studies have suggested substantial overlap in the etiology of CFS and FMS (Ablin et al., 2012; Van Houdenhove, Kempke, & Luyten, 2010; Van Houdenhove et al., 2013). However, it should be noted that not all CFS patients experience chronic pain, pointing to the need to identify subgroups of patients (Ablin et al., 2012; Kempke et al., 2013a). CFS is also associated with high rates of both lifetime and current psychiatric comorbidity, in particular depression (Arnold, 2008; Van Houdenhove & Luyten, 2006). The association between depression and CFS might be in part explained not only by shared risk factors (e.g. personality features such as perfectionism) (Luyten, Blatt, Van Houdenhove, & Corveleyn, 2006) but also by the fact that CFS itself is a risk factor for developing depression because of severe physical and mental impairments leading to social isolation and feelings of hopelessness (Arnold, 2008).

A Working Model of CFS

Although much remains to be learned about the causes of CFS, there is compelling evidence that CFS can best be conceptualized as a stress-related disorder with both psychosocial (i.e. personality and (early) life stress) and biological (i.e. neurobiological and genetic factors) factors playing a role in the etiology and maintenance of this disorder (Luyten, Kempke, Van Wambeke, Claes, Blatt, & Van Houdenhove 2011; Nater, Maloney, Heim, & Reeves, 2011; Van Houdenhove & Luyten, 2008; Van Houdenhove et al., 2013). Specifically, evidence over the past years suggests that CFS may reflect loss of resilience of the hypothalamic–pituitary–adrenal (HPA) axis, the main human stress response system, after a prolonged period of chronic stress (i.e. childhood adversity and/or cumulative life stress) or overexertion (Kempke, Luyten, Mayes, Van Houdenhove, & Claes, 2014; Luyten et al., 2011; Van Houdenhove & Luyten, 2010; Van Houdenhove et al., 2013; Van Houdenhove, Van Den Eede, & Luyten, 2009). The onset of the disorder is often triggered by

acute stressors, which can be either physical (e.g. after a surgery, an infection) or psychosocial (e.g. loss experiences) or a combination of both (Van Houdenhove & Egle, 2004; Van Houdenhove et al., 2010, 2013).

There is indeed increasing evidence from neurobiological research that in at least a subgroup of patients, CFS is associated with lower cortisol levels and blunted cortisol responses to stressors, suggesting reduced HPA axis functioning in these patients (Cleare, 2003; Papadopoulos & Cleare, 2011; Tak et al., 2011; Van Houdenhove et al., 2009). This is not only congruent with the recent emphasis on effort intolerance in the diagnosis of CFS (Carruthers et al., 2011) but also with the conceptualization of "allostatic" load in explaining stress-related conditions referring to the "wear and tear" of chronic stress on stress-regulating systems (McEwen, 1998, see also Arroll, 2013). This is also congruent with more general findings showing that chronic stress may eventually result in a "switch" (or neurobiological "crash") from hyperactivity to hypoactivity of the HPA axis (Fries, Hesse, Hellhammer, & Hellhammer, 2005; Lupien, McEwen, Gunnar, & Heim, 2009; Van Houdenhove et al., 2009).

The association between chronic stress and HPA axis dysregulation in CFS may be mediated by so-called epigenetic modifications (i.e. changes in the expression and activation) of genes that have been shown to regulate the neuroendocrine system (Van Houdenhove et al., 2013). Indeed, ground-breaking advances have recently been made in our understanding of the epigenetic regulation of the HPA axis (Bick et al., 2012), but studies in CFS are currently lacking. Importantly, the HPA axis has been shown to interact with other neurobiological systems such as neurotransmitter systems, the immune system, and pain processing systems, which may explain in part the high comorbidity between fatigue, pain, and depression (Van Houdenhove & Egle, 2004). For instance, reduced HPA axis function may be responsible for the typical "sickness response" (or flu-like feelings) of many CFS patients, since hypocortisolism may lead to low-grade immune activation (Silverman, Heim, Nater, Marques, & Sternberg, 2010).

In sum, evidence is growing that CFS may reflect a neurobiologically based loss of "adaptability" because of prolonged exposure to chronic stress or overexertion in psychologically and perhaps also genetically susceptible individuals (for a review, see Van Houdenhove et al., 2013).

Perfectionism as a Precipitating and Perpetuating Factor in CFS

Elsewhere, we have put forward a theoretical model in which increased vulnerability for chronic stress and subsequent neurobiological alterations in CFS may result, in part, from elevated levels of self-critical or maladaptive perfectionism (Kempke et al., 2013a; Kempke, Luyten, Mayes et al., 2014; Kempke et al., 2013c; Luyten et al., 2011; Van Houdenhove et al., 2013). This model is depicted graphically in Fig. 5.1. Of course, perfectionism is not the only factor contributing to chronic fa-

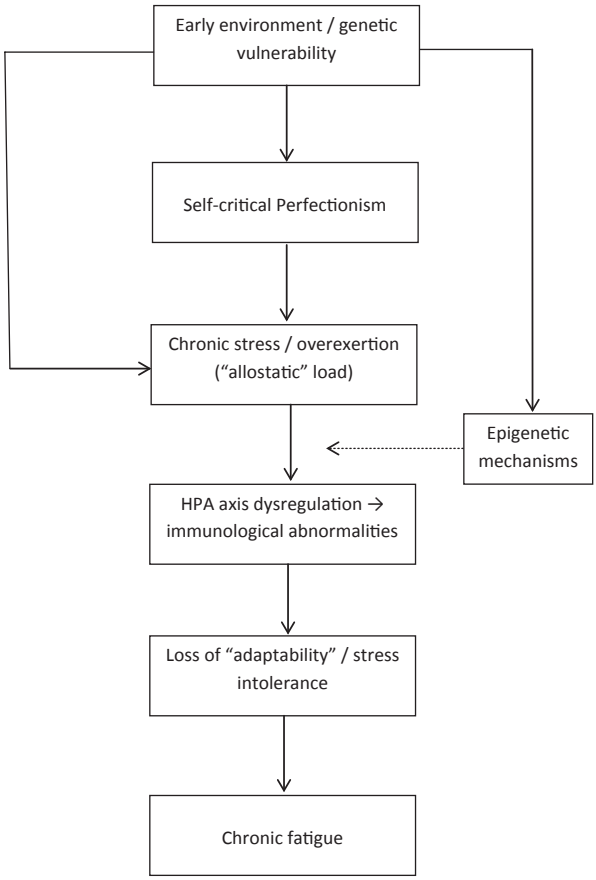

Fig 5.1 Role of perfectionism in the development of CFS

tigue conditions and certainly not involved in all cases with CFS (for an overview of risk factors see Van Houdenhove & Luyten, 2008). Further, congruent with the developmental psychopathology principle of multifinality (i.e. similar risk factors can lead to different outcomes) (Luyten, Vliegen, Van Houdenhove, & Blatt, 2008), it is clear that maladaptive perfectionism is a "transdiagnostic" vulnerability factor that is implicated in the development and maintenance of a wide range of negative outcomes (e.g. low self-esteem) and disorders, most notably affective spectrum disorders (Egan, Wade, & Shafran, 2011; Luyten & Blatt, 2013).

Clinicians and researchers have long theorized that perfectionistic attitudes are implicated in chronic fatigue and pain conditions. CFS patients have been described as overcommitted, conscientious, hard-working, overactive, and achievement oriented (van Geelen, Sinnema, Hermans, & Kuis, 2007; Van Houdenhove, Neerinckx, Onghena, Lysens, & Vertommen, 2001). Several theoretical models have been employed to measure perfectionism in CFS. Frost's two-factor model (Frost, Marten, Lahart, & Rosenblate, 1990) distinguishes between two dimensions of perfectionism, that is, adaptive or positive (i.e. normal achievement strivings) and mal-

adaptive or negative (i.e. maladaptive evaluative concerns (MEC)) perfectionism. From a psychodynamic perspective, Blatt's conceptualization of self-criticism or self-critical perfectionism (SCP) refers to a personality style characterized by both high personal standards and overly critical self-evaluations (Blatt, 2004; Kempke et al., 2013a). Numerous studies have shown that MEC or self-criticism is indeed associated with maladaptive outcomes (Stoeber & Otto, 2006; Dunkley, Blankstein, Masheb, & Grilo, 2006).

With a few exceptions (Blenkiron, Edwards, & Lynch, 1999; Wood & Wessely, 1999), studies have revealed a positive association between perfectionism and chronic fatigue. Magnusson et al. (1996) showed that maladaptive perfectionism was significantly associated with mental and physical trait fatigue among female nurses. In a large sample of CFS patients, Kempke et al. (2011a) found evidence for a two-factor model of perfectionism in CFS, that is, maladaptive, but not adaptive, perfectionism was positively related to severity of fatigue in CFS. In accordance with a study by Deary and Chalder (2010), analyses also indicated a substantial association between both perfectionism dimensions, suggesting that both factors are interrelated in CFS patients, that is, high achievement strivings are not really "adaptive" in these patients and are coupled with MEC (i.e. self-criticism). Indeed, Luyten and colleagues (2006) found that CFS patients retrospectively reported both higher levels of adaptive and maladaptive perfectionism before illness onset.

Further, case-control studies have shown significantly increased levels of unhealthy perfectionism in CFS patients. White and Schweitzer (2000), for instance, showed higher levels of maladaptive perfectionism among CFS patients compared to non-fatigued controls. Likewise, Deary and Chalder (2010) reported higher levels of maladaptive, but not adaptive, perfectionism among CFS patients compared to healthy controls. Recently, Sirois and Molnar (2014) replicated and extended previous findings by demonstrating higher levels of maladaptive perfectionism in CFS patients relative to well controls, patients with irritable bowel syndrome, and patients with FMS or arthritis. Moreover, CFS patients showed higher levels of self-blame coping compared to normal controls, congruent with findings on the specific role of self-criticism in these patients.

Prospective studies have provided further support for cross-sectional findings on the relationship between perfectionism and fatigue (Dittner, Rimes, & Thorpe, 2011; Kempke et al., 2013a; Moss-Morris, Spence, & Hou, 2010). Of particular note is a study conducted by Moss-Morris and colleagues (2010) showing that maladaptive perfectionism predicted the onset of CFS in patients with an acute episode of infectious mononucleosis (glandular fever). Dittner et al. (2011), in turn, showed that negative perfectionism significantly predicted fatigue levels in undergraduate students following a period of academic stress. Finally, a recent diary method study by our group (Kempke et al., 2013a) demonstrated that SCP predicted daily fatigue and pain symptoms in CFS patients over a 14-day period, attesting to its important role in the course of this disorder.

Although perfectionism seems to be more closely related to CFS, congruent with its transdiagnostic status and the high rate of comorbidity between functional somatic syndromes, it is important to note that unhealthy perfectionism and related

factors such as "counterdependency" (i.e. an overreliance on autonomy and self-sufficiency) and "all-or-nothing" behaviour (see below) have also been linked to chronic pain and gastrointestinal disorders such as irritable bowel syndrome and functional dyspepsia (Gregory, Manring, & Wade, 2005; Lerman, Rudich, & Shahar, 2010; Lerman, Shahar, & Rudich, 2011; Ochi et al., 2008; Spence & Moss-Morris, 2007; Rudich, Lerman, Gurevich, Weksler, & Shahar, 2008).

Finally, another important line of evidence for the role of perfectionism in CFS comes from research showing features of obsessive-compulsive personality disorder (OCPD) in CFS patients, a personality disorder that is mainly characterized by perfectionism and a strong need for control (Kempke et al., 2013c; Nater et al., 2010). For instance, we recently investigated personality features and personality disorders in CFS patients compared to well-matched control groups consisting of normal community individuals and psychiatric patients (Kempke et al., 2013c). As expected, CFS patients did not show an increased prevalence of personality disorders, but they did show elevated levels of obsessive-compulsive (and depressive) personality features, congruent with findings on the role of maladaptive perfectionism in CFS.

Perfectionism and Fatigue: Possible Mechanisms

The studies reviewed above clearly suggest a relationship between perfectionism and CFS. Importantly, research has also identified several potential mechanisms that may explain this relationship. Specifically, there is good evidence to suggest that perfectionism may be implicated in CFS because it is associated with (a) increased stress sensitivity, stress generation, and depression and (b) impairments in embodied mentalizing and emotion regulation leading to overexertion and other maladaptive behaviours that exacerbate and perpetuate symptoms. The common denominator of both pathways is that perfectionism overburdens the stress response system, thus contributing to "allostatic" load (Kempke et al., 2013a; Kempke, Luyten, Mayes et al., 2014).

Perfectionism, Stress Sensitivity/Stress Generation, and Depression

It has been shown that stress, and repeated or chronic stress in particular, may play a predisposing, precipitating (e.g. acute life stressors), and perpetuating role in CFS (Nater et al., 2011; Van Houdenhove & Luyten, 2008). There is increasing evidence to suggest that perfectionism, SCP in particular, is associated with chronic physical and/or mental stress and subsequent neurobiological alterations in CFS (Kempke, Luyten, Mayes et al., 2014; Van Houdenhove et al., 2013). Indeed, numerous studies have demonstrated that SCP is associated with both increased stress sensitivity and stress generation, making individuals vulnerable to stress-related conditions

(Blatt & Shahar, 2005; Luyten & Blatt, 2013). With regard to stress sensitivity, SCP has been related to both interpersonal- and achievement-related distress (i.e. holding excessive high standards for oneself leading to a sense of failure), suggesting that this personality dimension may reflect a general vulnerability factor to stress (Blatt & Shahar, 2005; Shahar, 2013).

Other studies have demonstrated that SCP is also associated with the "active" generation of stress, as indicated by findings that show that highly self-critical individuals experience more person-dependent (self-generated) stressors (e.g. negative interpersonal events) over time and tend to "degenerate" protective factors (e.g. social support) in their environment (Blatt & Shahar, 2005; Hammen, 2006; Luyten et al., 2006; Shahar & Porcerelli, 2006). Recently, our group (Luyten et al., 2011) replicated these findings in CFS patients by showing that SCP was associated with the generation of daily hassles over the last 2 months and with increased stress sensitivity over the course of a 14-day period. As a result, patients tended to show increased levels of depression, which has been shown to exacerbate fatigue symptoms and impede treatment response (Kempke et al., 2010; Kempke et al., 2011a). These findings are congruent with other studies showing that SCP in CFS may give rise to negative self-perceptions (i.e. lower self-esteem) and thus may increase vulnerability to depression (Kempke et al., 2011b; Kempke et al., 2011a).

Most recently, we provided evidence for the neurobiological mechanisms underlying the relationship between perfectionism and CFS. Specifically, we investigated the effect of SCP on cortisol stress response in CFS patients by using the Trier Social Stress Test (TSST) (Kirschbaum, Pirke, & Hellhammer, 1993), a widely used laboratory psychosocial stress induction paradigm involving a mathematics and public speech task in front of a video camera and an evaluative audience. Studies have shown that the TSST elicits a significant rise in cortisol (Kudielka & Wüst, 2010). CFS patients were instructed to provide saliva samples for cortisol analysis before the TSST, immediately after the test and at follow-up (i.e. 90 min relative to the onset of the TSST) to capture the complete stress process of each patient. In line with the hypotheses summarized above, we found that SCP was associated with higher subjective or self-reported stress, but with decreased cortisol stress reactivity after experimental stress induction and during recovery (Kempke, Luyten, Mayes et al., 2014). Thus, these results provided evidence that SCP may be one of the key factors that may contribute to reduced or blunted HPA axis reactivity in CFS, probably as a result of the "wear and tear" of prolonged chronic stress and/or overexertion, in combination with high subjective stress (Kempke, Luyten, Mayes et al., 2014a). Notwithstanding these promising results, further research is needed to unravel the neurobiological effects of SCP in CFS.

Perfectionism, Embodied Mentalizing/Emotion Regulation, and Overexertion

Perfectionism in CFS has also been associated with impairments in embodied mentalizing (i.e. the capacity to reflect on bodily sensations in terms of mental states)

and emotion regulation, probably within the context of insecure attachment, leading to overexertion and other unhelpful behaviours, thoughts (e.g. catastrophizing), and maladaptive coping strategies (Luyten, Van Houdenhove, Lemma, Target, & Fonagy, 2012, 2013), further exacerbated by the often unbearable feelings of pain, fatigue, and resulting helplessness and worthlessness.

In particular, studies have demonstrated that CFS patients tend to have negative beliefs about the experience and expression of emotions (e.g. experiencing negative emotions as a sign of weakness or fearing that others will negatively react to displays of emotions) (Rimes & Chalder, 2010), show a tendency to emotional suppression (Hambrook et al., 2011), and are less likely to interpret physical symptoms in terms of negative emotional states (Dendy, Cooper, & Sharpe, 2001), reflecting the dismissive or avoidant attachment pattern (i.e. denial of attachment needs and emotional experiences) observed in CFS patients with high levels of SCP (Luyten et al., 2012, 2013). Dismissive attachment strategies are typically activated when confronted with increased levels of stress and are considered to be exacerbated by the disorder itself (i.e. suffering from "unexplained" symptoms), leading to impaired emotion regulation (Luyten et al., 2012, 2013).

As a result, CFS patients tend to engage in excessive physical and/or mental activities as a way to cope with negative self-feelings in an attempt to prove self-worth (Luyten et al., 2012, 2013; Van Houdenhove, Neerinckx, Onghena et al., 2001). In this regard, more than a decade ago, Van Houdenhove et al. (2001) argued that the achievement-oriented lifestyle observed in the history of many CFS patients may serve as a coping strategy to maintain self-esteem and to prevent depression. Congruent with this assumption, Nater et al. (2006), in a population-based study, reported that escape/avoidance (i.e. behavioural efforts to escape or avoid a stressor) was the most prominent coping style in CFS patients. Harvey, Wadsworth, Wessely, and Hotopf (2008), using a large prospective birth cohort, found that overactivity (i.e. excessive levels of exercise) was associated with an increased risk for CFS. Similarly, high "action-proneness", "persistence", and "all-or-nothing" behaviour have been empirically linked to chronic fatigue (Fukuda et al., 2010; Moss-Morris et al., 2010; Van Campen et al., 2009; Van Houdenhove, Bruyninckx, & Luyten, 2006).

The concept of "action-proneness" was coined by Van Houdenhove, Onghena, Neerinckx, and Hellin (1995) to describe a tendency towards direct action in CFS (and chronic pain) patients that may increase the risk of overburdening. In a series of studies, Van Houdenhove and colleagues (Van Houdenhove et al., 2006; Van Houdenhove, Neerinckx, Onghena et al., 2001; Van Houdenhove et al., 1995) found that CFS patients described themselves retrospectively as more "action-prone" compared to normal controls. Moreover, they also found a strong agreement between the patients' self-report and the judgment by significant others regarding patients' premorbid lifestyle, ruling out the possibility that patients may have idealized their premorbid situation (Van Houdenhove, Neerinckx, Onghena et al., 2001). More recently, in a qualitative study, Pemberton and Cox (2014) identified a premorbid state of constant action among CFS patients. Moreover, the concepts of "doing nothing" and "rest" were associated with negative feelings and beliefs in patients. These findings are consistent with studies showing an association between CFS and "persistence" as measured with the Temperament and Character Inventory (TCI),

referring to the tendency of patients to persevere despite fatigue and frustration, pushing themselves past their limits (Fukuda et al.2010; Van Campen et al., 2009). Similarly, Moss-Morris et al. (2010) reported that "all-or-nothing" behaviour (i.e. an activity pattern characterized by cycles of overactivity and complete rest) is one of the most important predictors of developing CFS in patients with glandular fever.

Clinical experience and some empirical evidence suggest that CFS patients with high self-criticism are especially prone to "all-or-nothing" behaviour because of their inability to tolerate inactivity and rest (e.g. doing nothing is associated with feelings of guilt because of not being productive) (Kempke et al., 2013a; Luyten et al., 2011; Moss-Morris et al., 2010). Once ill self-critical patients may lower their excessive standards (Luyten et al., 2006), but may still show the tendency to exceed their mental and physical limits. Indeed, these patients often show periodic "outbursts of activity" when feeling somewhat better, followed by post-exertional malaise and complete rest, thereby exacerbating symptoms and impairments (Luyten et al., 2011; Luyten, Van Houdenhove, Pae, Kempke, & Van Wambeke, 2008; Moss-Morris et al., 2010). Although understandable as CFS can be completely incapacitating, in the long run, a "boom and bust" pattern increases the wear and tear on the stress response system (Luyten et al., 2012, 2013).

Origins of Perfectionism in CFS

Research suggests that SCP may be in part genetically determined (Moser, Slane, Alexandra Burt, & Klump, 2012) and first develops in the context of early attachment relationships (Blatt, 2004; Soenens, Vansteenkiste, & Luyten, 2010). Specifically, SCP appears to reflect an overemphasis or exaggeration of the development of the self and autonomy to the neglect of the development of the capacity for relatedness (Blatt, 2004; Luyten & Blatt, 2013) and may develop in an attempt to avoid rejection and criticism (or to seek approval from others) as a result of cold, critical, intrusive parenting characterized by conditional affection (i.e. parents make their affection conditional upon meeting high standards), and/or low emotion expressiveness (Blatt & Homann, 1992; Flett, Hewitt, Oliver, & Macdonald, 2002; Greenspon, 2008; Shahar & Henrich, 2013; Soenens et al., 2010). From this perspective, perfectionism in CFS may be a coping mechanism to compensate for feelings of worthlessness and inferiority (Kempke et al., 2013a; Kempke, Coppens, Luyten, & Van Houdenhove, in press; Van Houdenhove, Neerinckx, Onghena et al., 2001)—sometimes also reflected in excessive self-sacrificing tendencies (Van Houdenhove, Neerinckx, Onghena et al., 2001; Van Houdenhove et al., 2001; Hambrook et al., 2011).

Longitudinal research investigating these assumptions in CFS specifically is lacking, although there is evidence that CFS is related to a history of exposure to adverse childhood experiences (for a review, see Kempke et al., 2013b). For instance, a recent meta-analysis revealed a substantial relationship between self-reported psychological trauma and functional somatic syndromes, with the largest association in CFS (Afari et al., 2014). In a landmark study published in 2001, Van

Houdenhove et al. showed elevated levels of early adversity, in particular emotional trauma, in a mixed sample of CFS and fibromyalgia patients. They found that more than 60 % (i.e. 64.1 %) had a history of early adversity, with a considerable proportion reporting lifelong victimization (e.g. living in an abusive relationship). Similar results have been found in population-based studies conducted in the USA. Heim et al. (2006, 2009) showed that early trauma was associated with a three- to eight-fold increased risk for CFS. More recently, Kempke et al. (2013a) replicated and extended these findings in a sample of 90 well-screened CFS patients by showing that more than half of the patients (54.4 %) had experienced at least one type of early childhood adversity and more than 40 % multiple trauma, with the majority of these patients reporting emotional abuse and/or emotional neglect. Hence, CFS may be more closely associated with more subtle, "emotional" adverse childhood experiences rather than more "objective" or physical types of trauma, congruent with findings showing negative self-perceptions (i.e. high self-criticism) and a tendency towards overcompensation in these patients.

Clinically, CFS patients with a history of early adversity are particularly vulnerable to person-dependent stressors (e.g. daily hassles) and interpersonal difficulties, which may perpetuate and even exacerbate symptoms (Kempke et al., 2013b; Luyten et al., 2013). This is congruent with both animal and human studies suggesting that adversity, especially in early developmental stages, may have serious consequences for the developing stress system though its impact on brain structures and the epigenetic expression of genes involved in stress processing, leading to increased stress sensitivity, thereby increasing vulnerability to chronic stress and neuroendocrine dysregulation in later life (Lupien et al., 2009; Luyten et al., 2008; Uhrlass & Gibb, 2007; Van Houdenhove, Luyten, & Egle, 2009). For instance, Heim et al. (2009) found that only CFS patients with a history of early trauma showed reduced basal HPA axis activity. Recently, we demonstrated a significant association between emotional neglect and reduced HPA axis reactivity following experimental stress induction with the TSST paradigm (Kempke et al., 2015). Of note, similar results have been reported in patients with FMS (Riva, Mork, Westgaard, Ro, & Lundberg, 2010), suggesting that decreased HPA axis functioning might be implicated in both CFS and FMS and that these conditions are part of one larger spectrum of fatigue and pain conditions.

Conclusions and Clinical Implications

In this chapter, we provided evidence that self-critical or maladaptive perfectionism is implicated as a predisposing and perpetuating factor in at least a subgroup of CFS patients. Moreover, we summarized growing evidence suggesting that SCP is one of the key factors that may contribute to chronic stress and overexertion in CFS, thereby overburdening the stress response system. Finally, we discussed theory and research linking SCP to disruptions in early attachment relationships, congruent with studies demonstrating high rates of early childhood adversity in CFS patients.

Future research should concentrate on possible epigenetic mechanisms (i.e. DNA methylation) involved in the relationship between early adversity, SCP, and stress sensitivity in later life (Van Houdenhove et al., 2013), which is likely to further our understanding of the etiology of this condition and functional somatic syndromes in general. Furthermore, there is a pressing need for more longitudinal developmental studies concerning the origins of SCP, especially with regard to the emotional family environment and its interaction with genetic vulnerability including gene–environment interactions, in CFS and related conditions such as FMS (Kempke et al., 2013a; Kempke et al., 2015).

The findings reviewed in this chapter have important implications for the treatment of CFS. The most common treatment for chronic fatigue and pain conditions is cognitive behavioural therapy (CBT) and graded exercise therapy (GET) (Knoop, Prins, Moss-Morris, & Bleijenberg, 2010; Price, Mitchell, Tidy, & Hunot, 2008), although a number of other psychological treatments, including mindfulness-based treatment (Fjorback et al., 2013), acceptance and commitment therapy (Wicksell et al., 2013), and mentalization-based treatment (Luyten et al., 2012), have also been proposed. Regardless of theoretical orientation, research suggests that treatment of CFS should include a focus on perfectionism and related cognitive-behavioural factors, such as exaggerated "action-proneness" and "all-or-nothing" behaviour, in at least a subset of patients (Surawy, Hackmann, Hawton, & Sharpe, 1995; Van Houdenhove et al., 2006; Moss-Morris et al., 2010). For instance, Van Houdenhove and colleagues (2006) showed that high "action-proneness" in CFS could be changed by a multidisciplinary CBT-based group treatment. Likewise, Morlion et al. (2011) demonstrated significant changes in "action-proneness" in chronic pain patients who followed a brief cognitive-behaviourally based psycho-educational treatment.

Importantly, a number of studies on depression have shown that self-critical patients may negatively respond to brief, structured treatments because such interventions may undermine their sense of control and autonomy (Blatt, Zuroff, Hawley, & Auerbach, 2010; Luyten, Corveleyn, & Blatt 2005). Moreover, it has been shown that self-critical patients need more time to establish and maintain a therapeutic alliance and hence need more intensive treatment (Blatt et al., 2010; Luyten et al., 2005). Congruent with these findings, we recently showed that pretreatment SCP negatively predicted treatment outcome in terms of pain severity in patients with chronic pain following a brief, psycho-educational CBT-based treatment (Kempke, Luyten, Van Wambeke, Coppens, & Morlion, 2014). Also, perfectionism has been associated with lack of acceptance and the typical "boom-and-bust" activity pattern in CFS, leading to (periodic) overactivity and increased levels of depression, thereby impeding treatment response (Brooks, Rimes, & Chalder, 2011; Kempke et al., 2013a; Moss-Morris et al., 2010).

Hence, it is reasonable to suggest that CFS patients with high levels of SCP might benefit more from intensive and/or tailored treatment. In this regard, a wide range of specialized interventions exist to treat self-critical perfectionistic tendencies, including cognitive-behavioural and psychodynamic approaches (Blatt, 2004; Flett & Hewitt, 2008; Glover, Brown, Fairburn, & Shafran, 2007; Kannan &

Levitt, 2013; Luyten et al., 2012, 2013). In general, treatment for CFS patients with high levels of SCP should be aimed at helping patients change typical patterns of feeling and thinking associated with perfectionism (i.e. excessive need for the approval of others, negative beliefs about emotions) (Jason et al., 2013; Luyten & Van Houdenhove, 2013; Surawy et al., 1995; Van Houdenhove & Luyten, 2008; Van Houdenhove et al., 2013), which may facilitate behavioural and lifestyle changes.

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