Understanding self-experience in adolescent chronic fatigue syndrome

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Understanding self-experience in adolescent chronic fatigue syndrome

Het begrijpen van de zelf-ervaring in het
chronisch vermoeidheidssyndroom gedurende de adolescentie

(met een samenvatting in het Nederlands)

Proefschrift

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Searching the ox…

CONTENT

Introduction

> *"In order to be able to give a[n] explanation of how a condition of illness originated, a clarification, of course, is needed beforehand regarding what this condition of illness is in itself. […] What good is all explaining if what has to be explained remains unclear?"* (Heidegger, Zollikon Seminars[1])

The above quote seems to apply directly to the nosological class of illnesses known as the functional somatic syndromes. While the causes and pathogenesis of these conditions are the subject of constant discussion and controversy, the question of what kind of phenomena we are actually trying to explain tends to be neglected.

An ongoing debate

In June 2002, the Dutch Ministry of Health, Welfare and Sport asked the Health Council of the Netherlands -an independent scientific advisory body for the government and parliament on issues in the field of public health- to investigate the chronic fatigue syndrome (CFS). CFS, the main topic to be discussed in the subsequent chapters of this thesis, was regarded as a serious clinical condition leading to severe limitations in the physical, personal and social functioning of individuals afflicted by it. However, the syndrome seemed to be surrounded by uncertainty and dispute, as there was no consensus regarding its etiology, pathology or treatment. In addition, diagnostic tests for CFS were unavailable and the societal and economic costs were estimated to be up to 600 million euros per year[2]. The Health Council was therefore asked to assess the scientific state of affairs regarding the syndrome, focusing on questions pertaining to its definition, its possible causes, its course, its overlap with other conditions and diseases, and the potential therapeutic interventions, among others.

In January 2005, after more than two years of study, the special committee assigned with this task -chaired by former minister of Health, Welfare and Sport, Els Borst-Eilers and consisting of experts in the fields of internal medicine, general practice, psychoneuroimmunology, psychiatry, epidemiology and medical psychology- presented the new minister of Health, Welfare and Sport, Hans Hoogervorst, its extensive report on CFS[3]. The report contained conclusions concerning the syndrome's case definition, its incidence,

the role of the doctor-patient relationship, the range of factors influencing its etiology, the management of complaints, guidelines for patient care, the necessity of special attention for CFS in young people, the effectiveness of cognitive behavioral and graded exercise therapy in its treatment, the need for the further refinement of interventions and the requirement of a multidisciplinary approach in the scientific research into the condition. The main conclusion of the committee however, was that CFS should be considered *a genuine, severely incapacitating disorder*.

In February 2005, a mere 10 days later, the minister gave the President of the Dutch House of Representatives his response to the report. In this reaction, he indicated that -while he realized that individuals with CFS experienced severe complaints and difficulties- he was extremely hesitant to acknowledge the syndrome as a discrete and well-defined disease entity. If we were to do so, he argued, there would be no reason not to accept other medically unexplained conditions -such as fibromyalgia, irritable bowel syndrome and multiple chemical sensitivity for example- as independent disease entities as well, despite a clear lack of scientific evidence for such a conclusion. Furthermore, he doubted whether such recognition would be useful in clinical practice. He suggested that it would not only lead to inaccurate diagnoses, but also to incorrect treatments. Finally, he stated that – as the presumed main difference between CFS and ailments such as burn-out was the avowedly somatic attributions made by individuals with the former condition- the acknowledgement of CFS as a genuine disorder would have a negative influence on patients' willingness to seek health improvement. For, he argued, as patients with CFS believe in an exclusively physical cause of their symptoms, the acknowledgement of the syndrome as a genuine disease, would only strengthen their illness behavior and impair a self-initiated return to health.

Now, whether the minister was right with these inferences from, and interpretations of the Health Council's report on CFS is presently not the point. Heart of the matter is that his reaction once more sparked heated debates in the media among political parties, scientific researchers, patient groups, health care practitioners and the public at large. Ever since CFS-like conditions were first described in medical settings and brought to society's attention, the topics of discussion generally remained the same[4]; "Is it a real

disease?", "Is it physical?", "Is it mental?", "What causes it?", "Does it have a viral, or a bacteriological origin?", "Do the symptoms stem from depression, or emotional problems?", "Isn't everyone tired at times?", "Are patients just overstressed, and exaggerating their problems in order to avoid school or work?", "What are possible pharmacological solutions?", "Can psychotherapy cure the disorder?", "Is the effectiveness of alternative, non-conventional treatments for the condition deliberately downplayed?" and so on. In the general and scientific polemic almost any kind of negative or affirmative answer to these questions can be found. A definite resolution of these arguments seems unattainable, and it appears as if there is no common ground of agreement, or consensus with regard to any aspect of CFS. There is one thing however, virtually everyone –whether politician, researcher, patient, clinician, or lay person- agrees upon; whatever its cause, the experience of individuals with CFS of themselves as seriously fatigued and severely limited in bodily, emotional and relational regard is real and undeniable.

So far, research attention has primarily focused on investigations of etiology, pathology and treatment. Very few scientific studies however, have systematically asked the principal question; What kind of experiential phenomenon are we actually trying to explain? Related questions would be; Are fatigue and functional impairment truly at the core of patients' self-experience? In what way does their self-experience differ from that of healthy individuals? Does it differ from the self-experience of other chronically ill patients? Is their self-experience subject to change over the course of time? What methods should we employ to provide empirical answers to these questions? This thesis aims *a)* to further the understanding of self-experience in adolescent CFS by a thorough analysis of the affective self-narratives of teenagers with the syndrome, and *b)* to study whether narrative transformation –through counseling based on their self-experience- will contribute to an improved physical and psychosocial outcome.

Outline of the thesis

The research described in this thesis was conducted in three different studies. The first was a literature study reviewing personality in CFS. The second was a comparative, longitudinal investigation including adolescent patients with CFS and adolescent patients with juvenile idiopathic arthritis (JIA) -seen in the University Medical Center Utrecht, The Netherlands- and healthy controls. The third was a follow-up study of a previous cohort of adolescent patients with CFS –also seen in the University Medical Center Utrecht, The Netherlands. These studies resulted in the following chapters.

Chapter 1 is an introductory chapter, starting with a description of different aspects of CFS –its symptomatology, its overlap with other medically unexplained conditions, its prevalence and incidence, its prognosis, diagnostic criteria, possible etiologies and potential treatments. As (human) experience is always the experience of a particular person, a logical starting-point for a systematic investigation of self-experience in CFS is to study the personality of patients. Subsequently therefore, personality research in the syndrome is reviewed, and methodological and conceptual issues in these studies are discussed. Recommendations for the design of new empirical research are given.

Chapter 2 briefly describes dialogical self theory -on which this thesis' main empirical study is based. Grounding in the conceptual discussions and methodological recommendations from chapter 1, it cross-sectionally describes the self-positioning of adolescents with CFS, and compares it to the self-positioning of adolescent patients with JIA and healthy controls.

Chapter 3 introduces the selfconfrontation method (SCM) –the primary instrument used in the study. Going beyond the self-positioning described in chapter 2, it cross-sectionally describes the affective self-narratives of adolescents with CFS, and compares them to the affective self-narratives of adolescent patients with JIA and healthy controls.

Chapter 4 describes the long-term outcome of another cohort of adolescent patients with CFS. As the main study did not include a waiting list control group, adolescent patients who had previously participated in scientific studies into CFS in the same academic medical center were followed-up, in order to get a sense of the prognosis of teenagers with the syndrome after regular care.

Chapter 5 presents the use and effectiveness of the SCM as a self-management tool in adolescent CFS. It describes the transformation occurring in the self-experience of adolescents with CFS over a period of 4 months and seeks to relate these alterations to changes in health.

Chapter 6 is the general discussion, in which an attempt is made to integrate the empirical findings into an outline of a new conceptual framework for the understanding of self-experience in adolescent CFS.

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Chapter 1

Personality and chronic fatigue syndrome: Methodological & conceptual issues

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ABSTRACT

Objective: Among clinical psychologists, consulting physicians, scientific researchers and society in general an image has emerged of patients with chronic fatigue syndrome (CFS) as perfectionist, conscientious, hardworking, somewhat neurotic and introverted individuals with high personal standards, a great desire to be socially accepted and with a history of continuously pushing themselves past their limits. The aim of this article is to (a) give a concise review of the main recent studies on personality and CFS, (b) address the major methodological problems in the study of personality in CFS and (c) discuss some of the conceptual assumptions that seem to limit the research on personality and CFS. **Method:** A review study. **Results:** The results of the reviewed studies range from no evidence of major differences between the personalities of patients with CFS and controls, to evidence of severe psychopathology and personality disorder in patients with CFS. Although personality seems to play a role in CFS, it is difficult to draw general conclusions on the relation between personality and CFS. It is argued that this is partially due to the diversity and heterogeneity in study methods, patient populations, control groups and CFS case definitions. Personality should be regarded as an important factor to be studied in CFS. However, additional studies are needed, not focusing exclusively on personality *disorder*, or personality considered on a general trait level. **Conclusions:** In recent developments in personality research, the continually evolving life narrative that makes sense of, and gives direction to, an individual's life is also regarded as an important aspect of personality. New insights into personality and CFS might be gained by systematically studying the self-narratives of patients with the syndrome.

INTRODUCTION

This article's main concern is the study of personality in the chronic fatigue syndrome (CFS). CFS is a syndrome of unknown origin. It is mainly characterized by a severely disabling fatigue and it is commonly associated with symptoms such as myalgias, headache, sleep disturbance, swollen lymph nodes and cognitive impairment. In recent years CFS has become a growing concern, not only for patients suffering from the illness and for their families, but also for medical science, clinical psychology and society in general.

Some of these concerns already become apparent in defining what CFS is. In many cases, it is difficult to distinguish between idiopathic chronic fatigue, CFS and other unexplained medical conditions such as fibromyalgia, tension headache and irritable bowel syndrome, as these seem to be very similar and substantially overlapping[1,2]. As no causes for CFS are found and definite markers for the illness are absent, the diagnostic process is usually extended and patients have to go through a whole battery of laboratory tests, physical examinations and psychological investigations before they are diagnosed with CFS. In 1988, the US Centers of Disease Control (CDC) proposed a set of diagnostic criteria to facilitate scientific research into CFS[3]. However, these criteria were criticized, not only because a large number of symptoms had to be present for a diagnosis of CFS, which might bias in favor of psychiatric morbidity[4], but also because it excluded such conditions as anxiety and depression, which some propose to be a result of the syndrome[5,6]. Therefore, less restrictive criteria were developed, amongst others in the UK by Sharpe et al.[7]. Consequently, the CDC criteria were also revised[8]. At present these criteria are generally accepted and used for international research purposes. In table 1, these criteria are presented.

Estimations on the prevalence of CFS range from 37/100 000[9], to 75-267/ 100 000[10], and even 740/ 100 000[11]. However, these numbers are difficult to compare as different populations were studied and varying CFS case definitions were used. Estimations on the incidence of CFS are rare but, based on their assumptions with regard to the prevalence of CFS, Lawrie et al.[11] estimated the annual incidence of CFS to be 370/ 100 000.

Full recovery from CFS is unusual. In a recent review[12] of studies on the prognosis of CFS, it was found that the median full recovery rate was only 5% and the median proportion of patients who had improved

Table 1
US Centers for Disease Control Case Definition of CFS, 1994[8]

Diagnostic criteria:

At least 6 months of persistent or recurring fatigue for which no physical explanation has been found and which

- is of new onset, that is to say it has not been lifelong
- is not the result of ongoing exertion
- is not substantially alleviated by rest
- severely limits functioning

In combination with four or more of the following symptoms, persistent or regularly recurring over a period of six months and which must not have predated the fatigue:

- self-reported impairment in memory or concentration
- sore throat
- tender cervical lymph nodes
- muscle pain
- multi-joint pains
- headache
- unrefreshing sleep
- post-exertional malaise lasting 24 hours or longer

Exclusionary criteria:

- any medical condition that may explain the presence of chronic fatigue
- a psychotic, major or bipolar depressive disorder (but not an uncomplicated depression)
- dementia
- anorexia or bulimia nervosa
- alcohol abuse or the use of drugs
- severe obesity

during follow-up was 39.5%. The prognosis for children and adolescents however, is generally somewhat better[13]. In a recent follow-up study of adolescent patients with CFS[14] it was found that, at a mean of 4.57 years after initial examination, 25% of the patients showed near to complete improvement and 31% showed partial improvement.

Etiological studies into the possible causes of CFS have been abundant. Active viral infection has frequently been associated with the symptoms of CFS, but evidence for this hypothesis has not consistently been found. There appear to be no significant differences between patients with CFS and healthy controls in the prevalence of human herpes viruses, Epstein-Barr virus, cytomegalovirus, hepatitis C virus, adenovirus and parvovirus B19, amongst many others[15-17].

Immune dysfunction is another possible etiological factor that has been widely studied. Chronic lymphocyte overactivation with cytokine abnormalities in patients with CFS, associations between T cell markers and CFS, and associations between low natural killer cells and CFS have all been reported[18,19]. However, in a recent systematic review of the immunology of CFS, the authors noted that studies supporting almost any conclusion regarding the presence, or absence of immunological abnormalities in CFS could now be found, and concluded that no consistent pattern could be identified[20].

The same holds true of studies on the role of the neuroendocrine system in CFS. Disturbed neuroendocrine-immune system interactions, low circulating cortisol, high nocturnal melatonin, abnormalities in the relationship between cortisol and central neurotransmitter function, a disturbance of neurotransmitters in HPA axis function, and alterations in adrenal function in CFS have all been suggested and some evidence for these claims has been found[21-25]. Again however, in an extensive review on the neuroendocrinology of CFS, it was concluded that no consistent evidence of abnormalities could be found and that it was unclear whether neuroendocrine changes (if any) are primary or secondary to behavioral changes in sleep or exercise[26].

Along other lines of research, the psychiatric status of patients with CFS has received much attention. Several studies have reported a high prevalence of current psychiatric disorders in CFS, predominantly depression, somatization disorder and hypochondria[27,28]. However, while some studies concluded that psychiatric illness in many cases predated the development of CFS[29,30], other studies concluded that psychiatric disorder was concurrent with the onset of CFS and therefore more likely to be a consequence of, rather than a risk factor to CFS[31,32]. In that case, CFS is not seen as a manifestation of an underlying psychiatric disorder and more somatic causes are presumed[33].

Neuropsychological deficits and impaired cognitive functioning in patients with CFS have also received widespread attention, and have frequently been implied to be an important explanatory factor for some of the symptoms of CFS. People with CFS often complain of difficulties with memory and concentration. Several studies have described an impaired cognitive performance of patients with CFS on neuropsychological tests measuring speed of information processing, memory, motor speed and executive functioning[34,35]. Problems with neuropsychological functioning were found to be unrelated to depression, fatigue or anxiety[36] and have instead been related to low levels of physical activity[37], a more extensive use of frontal and parietal brain regions[38] and even genetic traits[39]. In contrast with this, many other studies have found no difference in cognitive performance between patients with CFS and controls, and no evidence of any neuropsychological deficits in CFS[40,41]. However, although in many studies objectively no cognitive differences between patients with CFS and controls are found, patients with

CFS consistently report cognitive complaints and underestimate their actual performance on neuropsychological tests. This difference between the subjective perception of cognitive impairment and the absence of any objective evidence has led some researchers to speculate that, in contrast to laboratory cognitive tests, in CFS everyday cognitive tasks may require excessive processing resources leaving patients with CFS diminished spare attentional capacity[42], and other researchers to suggest that patients with CFS set impossibly high standards of personal performance[43].

Other risk factors for the development of CFS that have been implied (and for which some evidence has been found) are birth order[44], family reinforcement of illness behavior[45], maternal overprotection in relation to the formation of belief systems about activity avoidance[46] and a family history of physical and mental illness[47]. However, as with all of the etiological studies that have been discussed so far, the contrary conclusions can also be found. In a large birth cohort study into childhood predictors of CFS in adulthood, in which more than 11 000 people were followed up until the age of 30, no associations between maternal or child psychological distress, parental illness or birth order, and an increased risk of lifetime CFS were identified[48].

There have also been many studies into possibly effective treatment strategies for CFS. However, presently there is no established, universally beneficial intervention for the management and treatment of CFS[49]. With regard to medical and pharmacological treatment, amongst others, intramuscular dialyzable leukocyte extract[50], intravenous immunoglobulin[51], hydrocortisone[52] and antidepressants[53,54] were investigated in placebo-controlled studies, without proving their effectiveness. Recently, the effects of galantamine hydrobromide[55], polynutrient supplements[56], homeopathic treatment[57] and corticosteroids[58] have been studied in randomized controlled trials, but were also found to be ineffective. At the moment, only cognitive behavior therapy[59-61] and graded exercise therapy[62,63] have shown some effectiveness, for a proportion of patients, in randomized controlled trials.

So, CFS seems surrounded by controversy. Patients are confronted with a highly ambiguous illness that severely incapacitates them. In addition to this they suffer from the consequences of the unclear medical status of the disease. Due to the uncertainties surrounding the etiology of CFS, its symptomatology and the overall objective 'realness' of the syndrome, they are likely to encounter disbelief concerning their medical condition[64]. At present it is being discussed whether the impact of labeling patients with a diagnosis of CFS is enabling, or rather disabling[65]. In the absence of a clear biological marker for the illness, which would permit a definite diagnosis instead of a descriptive one, based almost solely on the exclusion of other disease entities, patients are often faced with skepticism by their families, employers, insurance companies, psychologists and physicians. In a recent study on illness

experience in CFS it was found that lack of illness recognition ranked high as a source of dissatisfaction for patients and was thought to aggravate psychiatric morbidity[66]. In contrast with this, physicians participating in a study on their perspectives on patients with CFS[67] expressed the view that patients seem to exaggerate the severity of their problems, and that there appears to be a discrepancy between their reported health and the way they look and behave.

Although it has been widely recognized that a positive and co-operative caregiver-patient relationship is of the utmost importance in the successful treatment of CFS[68], uncertainty and conflicts about the causal attribution of the syndrome, in many cases, put this relationship under pressure. Steven et al.[69] showed that one-third of a group of more than two-thousand general practitioners did not believe that CFS was a distinct syndrome and thought the most likely cause was depression. This finding was confirmed by another study in which it became clear that while most of the doctors participating in the study believed CFS to have a psychological cause, all of the patients attributed their illness to a physical cause[70]. This disagreement over the perceived origins of CFS was thought to largely account for the fact that two-thirds of the patients in this study were dissatisfied with the quality of the medical care they had received.

This same dispute about the etiology of the syndrome, in combination with concerns about its nosological status, seems to have characterized and dichotomized medical and psychological thought on CFS. In spite of the great advances medical science has made in the explanation and treatment of diseases with an evident organic cause, the causes for CFS remain unclear and our understanding of the illness progresses only slowly. This *"prototypical mind/ body problem"* (p. 258)[71] seems to confront medicine with the limitations of the traditional paradigm, through which it has made such progress in the understanding and treatment of 'classical' diseases. As is now widely acknowledged the debates on chronic fatigue and immune dysfunction syndrome, neurasthenia, postviral fatigue syndrome, myalgic encephalomyelitis, chronic mononucleosis and chronic Epstein-Barr virus infection, as CFS was formerly known, were, and not uncommonly still are, characterized by a mind/ body dualism that seems inherent to a biomedical model of thought, oriented towards monocausal explanation[72-74]. On the one hand, there are those who believe that CFS is initiated by a still unknown physical cause such as a chronic or relapsing viral infection, immunological deficiencies or abnormalities in the neuroendocrinological system. The absence of a clear and objective organic cause, on the other hand, leads others to relegate CFS to the realm of the mental and 'subjective' illnesses. In that case CFS is mostly thought of as a psychiatric disorder (e.g. a masked expression of depression, or a form of somatization), or a cognitive phenomenon.

However, a more logical explanation of the variety of findings and opinions on CFS would be that the illness is multi-factorial. Social, mental and somatic causes, and psychological and physical effects are not easily discernible, but instead appear to be interrelated. In recent years, a more biopsychosocial approach in the scientific research into CFS has become the standard[75]. In line with this approach (and in addition to the already mentioned studies) researchers have now also begun to study the iatrogenic factors in CFS[70], associations in symptoms between patients with CFS and their parents[76], the illness beliefs and attributions of patients with CFS[77,78], the psychological adjustment of patients with CFS[79], the health-related quality of life of patients with CFS[80], the locus of health control in patients with CFS[81], the relationship between ethnicity and CFS[82], the coping strategies of patients with CFS[83], the influence of family members in CFS [84], the cultural and historical context of CFS[74, 85-88] and the personalities of individuals who have developed CFS (reviewed in this article).

So, within the biopsychosocial model of CFS one of the aspects studied, that might have a perpetuating and even a predisposing role in the syndrome, is the personality of people suffering from CFS. Among clinical psychologists, consulting physicians, scientific researchers and in society in general, a typical image has emerged of patients with CFS as perfectionist, conscientious, hardworking, somewhat neurotic and introverted individuals with high personal standards, a great desire to be socially accepted and with a history of continuously pushing themselves past their limits[89,90]. In addition to this, they are characterized as being particularly averse to any psychological or psychiatric explanation of the syndrome and extremely persistent in fixed beliefs concerning their illness, thereby reducing the chance of successful treatment[61]. However, this image of people suffering from CFS was never really scrutinized, with most of the research activity concerning the individual with CFS focusing on psychopathology and possible psychiatric disorder.

The aim of this article is to (a) give a concise review of the main recent studies on personality and CFS, (b) address the major methodological problems in the study of personality in CFS and (c) discuss some of the conceptual assumptions that seem to limit the research on personality and CFS.

METHOD

The PubMed and PsychINFO databases from 1988 (when the original Centers for Disease Control criteria for CFS were first established) to November 2006 were searched using the keywords *CFS and personality, CFS and psychology, CFS and individual, CFS and identity*. On PubMed this generated 623 hits and on PsychINFO an additional 333 hits. All 956 abstracts were read. In addition the reference lists of the retrieved articles were examined.

The intention in the selection of studies was to include all original articles describing primary research on personality and CFS. Review articles, articles describing studies without mentioning which CFS case definition criteria were used, or without an appropriate control group, and articles focusing exclusively on psychiatric morbidity, were all excluded. Using these criteria led to the inclusion of a final 16 studies*. This review might not have captured all relevant studies. However, the discussed articles are the most important ones and can be seen as representative of the current state of affairs in the field.

RESULTS

In table 2, a concise overview of the main recent studies on the role of personality in CFS is given. Studying these results, it soon becomes obvious that the findings regarding the association of personality and CFS are not definitive. Although some studies seem to confirm, for a proportion of patients, *some* of the aspects of the aforementioned stereotype of people suffering from CFS, other studies found no such evidence. Some findings however, seem to be more consistent than others.

All in all, there seems to be most empirical evidence for an increased level of neuroticism in patients with CFS. Taileffer et al.[93] found significantly higher neuroticism scores in patients with CFS compared to the general population. Chubb et al.[100] found increased scores in their CFS subjects with concurrent depression. Masuda et al.[94] found elevated neuroticism scores in their noninfectious CFS group, although not in their postinfectious CFS group. Fiedler et al.[99], Blakely et al.[107], Buckley et al.[101] and Johnson et al.[106] also found significant differences in neuroticism scores between patients with CFS and healthy controls and Rangel et al.[98] found the related items of conscientiousness, worthlessness and emotional lability to be significantly more common in patients than in controls. However, most subjects in their study were recovered and their mothers, instead of the patients themselves, had been used as informants. Several other important limitations in the interpretation of these findings regarding neuroticism should also be mentioned. One study found elevated scores of neuroticism only in comparison to non-study recruited norm values for a general population[93]. In addition, generally no differences in neuroticism between patients with CFS and other patients suffering from a chronic disease were found[93,103,106]. Other studies used the MMPI to detect neuroticism[105,107] which, due to its sensitivity to physical symptoms, has been found to perform poorly in CFS and to overestimate psychopathology in chronically ill populations[106] and finally, many of the findings of high neuroticism were later accounted for by co-morbid depression[93,99,100,106].

Table 2
Primary research on personality in CFS

Study	No. of Participants	Study Methods	Major Findings
Henderson and Tannock[92]	61 patients with CFS (CDC, 1994)[8] 40 psychiatric inpatients with depressive disorder 45 healthy controls	Structured Clinical Interview for DSM-III-R Diagnoses (SCID-II)	39% of the CFS group, 73% of the depressed group and 4% of the healthy group were diagnosed with personality disorders. Cluster C disorders (avoidant, dependent, obsessive-compulsive, self-defeating and passive-aggressive) were the most common in both the CFS and depressed group. Personality disorder in patient with CFS could not be accounted for by comorbid depression.
Taileffer et al.[93]	45 patients with CFS (CDC, 1988)[3] 40 patients with multiple sclerosis	Illness Worry Scale, Neo Five-Factor Inventory (NEO-FFI), SCL-90R Depression Scale, Symptom Interpretation Questionnaire (SIQ)	There was no difference between the groups on neuroticism, depressive symptoms, or on the SIQ. The CFS group did have significantly higher scores than the MS group on the Illness Worry Scale. When the CFS group was divided into more and less depressed patients, the neuroticism scores were found to be significantly higher than the general population in the more depressed CFS group.
Masuda et al.[94]	16 patients with postinfectious CFS (CDC, 1992)[95] 20 patients with noninfectious CFS (CDC, 1992)[95] 20 healthy controls	Holmes Social Readjustment Rating Scale, Cornell Medical Index (CMI), Maudsley Personality Inventory (MPI), Yatabe-Guilford test, Self-rating Depression Scale (SDS)	The stress, maladjustment, marked anxiety, depressive tendency and hypertense state scores of both CFS groups were significantly higher than in the control group. No significant differences between both CFS groups on these scores were observed. However members of the postinfectious CFS group were diagnosed as social extroverts, while those in the noninfectious CFS group were neurotic and introspective.
Van Houdenhove et al.[96]	A randomized sample of a 100 patients out of 124 patients with CFS (CDC, 1994)[8] 68 patients with fibromyalgia (FM)	Questionnaire for Habitual Action-proneness (HAB)	The patients and their significant others scored the questionnaire similar. These scores were higher than the norm values, suggesting that high 'action-proneness' and an associated 'overactive' lifestyle may be one of the factors playing a predisposing, initiating as well as a perpetuating role.

White and Schweitzer[97]	44 patients with CFS (CDC, 1994)[8] 44 healthy controls	Multidimensional Perfectionism Scale (MPS), Rosenberg Self-Esteem Scale (RSE), Courtauld Emotional Scale (CECS), Marlowe-Crowne Social Desirability Scale (MCS)	The study demonstrated higher perfectionism scores and lower self-esteem in individuals with CFS, than in individuals in the healthy control group. The results suggest that individuals with CFS have a maladaptive perfectionist personality style.
Rangel et al.[98]	25 adolescent patients with CFS (Oxford Criteria, 1991)[7] *At the time of the study two-thirds (n=17) had recovered and the subject's mothers were used as informants.* 15 healthy controls	Personality Assessment Schedule (PAS), Kiddie-SADS Psychiatric Interview (K-SADS), Children's Global Assessment Scale (CGAS), Child Behaviour Checklist (CBCL)	Subjects with CFS demonstrated increased scores for introspection, sensitivity, conscientiousness, vulnerability, lability and worthlessness. Personality difficulty may either be a contributory factor to CFS in children, or result from the prolonged disease.
Fiedler et al.[99]	35 veterans with CFS (CDC, 1994)[8] and co-morbid psychiatric disorder 23 veterans with CFS and no co-morbid psychiatric disorder 45 healthy veterans	Combat Exposure Scale (CES), Operation Desert Storm Survey (ODS Survey), Childhood Traumatic Events Scale, Psychiatric Epidemiology Research Interview-Life Events Scale (PERI), Neuroticism, Extroversion, Openness Personality Inventory (NEO-PI), Toronto Alexithymia Scale (TAS), Marlowe-Crowne Social Desirability Scale	Measures of personality and negative coping strategies (as well as self-reported combat and chemical exposures) significantly differentiated healthy veterans from those with CFS. On the neuroticism subscales of anxiety, hostility, depression, self-consciousness, impulsivity and vulnerability the CFS/ psychiatric group scored significantly higher than the two other groups. Veterans with CFS reported a poorer ability to identify and communicate feelings than did healthy controls.

Chubb et al.[100]	62 patients with CFS (CDC, 1994)[8] and 48 healthy controls completed the EPQ. 50 patients with CFS (CDC, 1994)[8], 100 healthy controls and 37 depressed patients completed ASQ.	Eysenck Personality Questionnaire (EPQ), Attributional Style Questionnaire (ASQ)	Patients with CFS and concurrent depression scored significantly higher than individuals with CFS without concurrent depression or healthy controls on the neuroticism subscale. On the social desirability subscale subjects with CFS did not differ from the controls. Scores on both questionnaires show no difference between patients with CFS and healthy controls except for those subjects with CFS who are also concurrently depressed. In these cases the scores resemble patients with depression.
Buckley et al.[101]	30 non-depressed patients with CFS (CDC, 1994)[8] 20 patients with major depressive disorder (MDD) 15 healthy controls	Revised NEO Five-Factor Inventory, Eysenk Personality Questionnaire	Higher scores on neuroticism and introversion in patients with CFS than in healthy controls. Lower neuroticism in CFS than MDD patients. Patients with CFS reported increased postmorbid neuroticism and introversion, suggesting that personality may have changed as a result of the illness.
Christodoulou et al.[102]	38 patients with CFS (CDC,1994)[8] 40 patients with multiple sclerosis 40 healthy controls	Diagnostic Interview Schedule (Q-DIS), Tridimensional Personality Questionnaire (TPQ)	Personality profiles of CFS and MS subjects were generally similar. Both the MS and the CFS groups showed elevated levels of Harm Avoidance and lower levels of Reward Dependence in comparison to healthy subjects. The only difference was on the dimension of persistence, where the CFS group displayed preserved persistence and the MS group showed a reduction. There was no evidence to suggest that patients with CFS possessed an unusual level of negativity that would have predisposed them to develop their illness.

Wood and Wessely[103]	101 patients with CFS (Oxford Criteria, 1991 and CDC, 1994)[7,8] 45 patients with rheumatoid arthritis (RA)	MacLean's questionnaire on attitudes towards mental illness, Social Desirability Questionnaire, Defensiveness Scale of Adjective Check List, Twenty-Item Toronto Alexithymia Scale, Tridimensional Personality Questionnaire, Multidimensional Perfectionism Scale, Beck Depression Inventory (BDI) Social Adjustment Scale (SAS)	Alexithymia scores were greater in the RA patient group and social adjustment was poorer in the CFS group. No differences were found between CFS and RA patients in measures of perfectionism, attitudes towards mental illness, defensiveness, social desirability, or sensitivity to punishment. There was no evidence from this study of major differences between the personalities of patients with CFS and patients with RA.
Blenkiron et al.[104]	40 patients with CFS (CDC, 1994)[8] 31 healthy controls	Multidimensional Perfectionism Scale, Chalder Fatigue Questionnaire, Hospital Anxiety and Depression Scale (HAD)	Women more than men with CFS tend to set lower expectations and standards for others. The values for perfectionism found on the MPS were lower in the CFS sample (reflecting fewer perfectionist traits) than in the control group. This may indicate that the CFS respondents in this survey had already moderated their perfectionist tendencies and reset their standards to cope with the unpredictabilities of the disorder.
Schmaling and Jones[105]	53 patients with CFS (CDC, 1988/ 1994)[3,8] 43 healthy controls	Minnesota Multiphasic Personality Inventory (MMPI)	The aggregate MMPI profile of patients with CFS suggests that they have significant physical complaints and difficulties with cognitive functioning, are concerned about their symptoms, and are emotionally distressed. Their profile is similar to that of patients with chronic pain.

Johnson et al.[106]	35 patients with CFS (CDC, 1988/ 1992)[3,95] 20 patients with multiple sclerosis 24 depressed patients 40 healthy controls	The NEO Neuroticism Scale, Personality Diagnostic Questionnaire-Revised (PDQ-R), Beck Depression Inventory	The study found progressively higher rates of personality disorders (PD) and neuroticism from healthy controls through CFS and MS (who did not differ) to the depressed group. The most common PD's among subjects with CFS were histrionic (23%) and borderline (17%). The CFS group with concurrent depressive disorder (34% of the CFS group) was found to account for most of the personality disorder.
Blakely et al.[107]	58 patients with CFS (McKenzie (New Zealand) criteria, 1988)[108] 81 patients with chronic pain (CP) 104 healthy controls	Minnesota Multiphasic Personality Inventory, Beck Depression Inventory, General Health Questionnaire (GHQ) Lazarus ways of Coping (WoC)	Progressively more elevated scores on most scales from healthy controls through chronic pain to patients with CFS were found. The individuals with CFS showed more deviant personality traits reflecting emotionality or neuroticism, inward hostility, self-criticism and guilt, although personality profiles fell into different groups. The hypothesis is brought forward that in CFS we are dealing with a particular subpopulation of patients with CP, who are particularly extreme and relatively homogenous in their endorsement of CFS symptoms.
Millon et al.[91]	24 patients with CFS (CDC, 1988)[3] No appropriate control group	Millon Clinical Multiaxial Inventory (MCMI-II), Profile of Mood States Hamilton Rating Scale of Depression (HAM-D), Folstein Mini-Mental Examination The Wechsler Memory Scale (WMS)	Evidence of severe personality pathology and affective distress was found. Anxiety, somatic disorder and depression were particularly prominent. Histrionic (33%), schizoid (29%) and avoidant, narcissistic and aggressive/ sadistic (each 25%) personality scales were pathologically elevated.

Furthermore, there also seems to be evidence for the prevalence of personality disorder in a *proportion* of patients with CFS. In the first study on personality and CFS, Millon et al.[91] found elevated base rate
means, above those of a non-clinical population, on the histrionic, schizoid and avoidant scales of the MCMI, measuring DSM axis II personality disorders. Henderson and Tannock[92] also found quite a high level of personality disorder (39%), predominantly obsessive-compulsive personality disorders, in their sample of patients with CFS. Similar rates and findings were reported by Ciccone et al.[27]. In the study by Johnson et al.[106], 37% of the subjects with CFS met the criteria for at least one personality disorder, predominantly histrionic and borderline personality disorders. So, there certainly seems to be a somewhat higher rate of personality disorder within the CFS population than in non-clinical populations, in which it is estimated to be between 10-19%[109,110]. However, personality disorder rates were similar in patients with CFS and those with other medical conditions[106]. Also, it should be noted that personality disorder was *not* found in the majority of patients. Furthermore, again there are some important confounding aspects and the generalizability of the findings in the abovementioned studies can be questioned. For example, some studies did not have a control group[27,91]. Moreover, the MCMI that Millon et al.[91] used includes many items that tap somatic concerns, thereby increasing the likelihood of a diagnosis of personality disorder in chronically ill patients. Co-morbid depression accounted for most personality pathology in one study[106] and although this was not the case in the study by Henderson and Tannock[92], they only included patients attending a teaching hospital, who are likely to have a more severe form of CFS.

Although perfectionism, social desirability and introversion have commonly been referred to as some of the most characteristic features of the personalities of patients with CFS, the scientific evidence on this subject is far less clear-cut. White and Schweitzer[97] found higher perfectionism scores in individuals with CFS than in their control group and Christodoulou et al.[102] found the only difference between their CFS and MS groups to be an elevated persistence score, which they related to perfectionism. However, in contrast to these findings Wood and Wessely[103], and Blenkiron et al.[104] did not find higher perfectionism scores in patients than in controls. There were three studies that specifically studied social desirability among patients with CFS[100,101,103], but these studies revealed no differences between patients and control groups. With regard to extroversion and introversion, Masuda et al.[94] found the members of their postinfectious CFS group to score higher on extroversion than controls, although the members of their noninfectious CFS group were found to be more introspective. And finally, while Buckley et al.[101] found that patients with CFS scored significantly lower than their healthy controls on

extroversion, Chubb et al.[100] found the scores on extroversion of their CFS group not to be different from those of their healthy control group.

So, the results vary from the uncovering of *"evidence of severe personality pathology and affective distress"* (p. 131)[91], to the finding of *"little evidence that any particular personality trait discriminates CFS patients […] from other patients suffering a physically disabling condition"* (p. 395)[103]. However, even when evidence of abnormalities in the personality profiles of patients with CFS is found, there remains a considerable lack of clarity regarding the precise role of personality in the syndrome, and this is reflected in the conclusions these studies draw. For example, while Van Houdenhove et al.[96] conclude that *"high 'action-proneness' and an associated 'overactive' lifestyle may be one of the factors playing a predisposing, initiating as well as a perpetuating role in CFS"* (p. 575), Christodoulou et al.[102] found no evidence to suggest that patients with CFS had any particular personality traits that would have *predisposed* them to develop their illness. Rangel et al.[98] conclude that personality difficulty might *either* be a contributory factor to CFS, *or* result from the prolonged disease, and Buckley et al.[101] and Blenkiron et al.[104] conclude that the personality of subjects with CFS *might* have changed as a result of their disease.

Although the impression of many psychologists, physicians and researchers, that the personality of patients is a factor in CFS, seems to be justified by clinical experience and is supported somewhat by the available research, decisive conclusions on this subject are difficult to draw on the basis of the relevant scientific studies. Even though these studies have scrutinized the aforementioned image of the 'typical' individual with CFS, no definitive conclusions for the patients as group can be drawn, and a general and uniform answer to the question of the role of personality in CFS is hard to formulate. A provisional conclusion might be that it is *"difficult to disentangle personality factors that may have contributed to the development of the condition from emotional reactions that are consequences of the debilitating symptoms and the mixed responses of others to the illness"* (p. 237)[72].

However, part of the reason for this opaqueness, seems to be due to a certain heterogeneity of the reviewed studies with regard to study methods, patient populations, control groups and CFS case definitions. Therefore, before discussing what seem to be some shared conceptual assumptions of these studies, in the next section some of the major methodological issues concerning the study of personality in CFS will be addressed.

DISCUSSION

Methodological issues regarding the study of personality in CFS

An obvious reason for the discrepancies in the conclusions of the studies discussed might be the use of different methods to measure personality. This

diversity seems almost inevitable when we consider the variety and divergence in health care settings and traditions of personality research. However, even when using the same instruments there often was no uniformity in the findings. In three studies, all using the Multidimensional Perfectionism Scale (MPS) for example, a remarkable lack of consensus in the results emerges. While White and Schweitzer[97] demonstrated *higher* perfectionism scores in individuals with CFS than in persons in their healthy control group, Wood and Wessely[103] using the same MPS, found *no differences* in measures of perfectionism between the patients with CFS and the patients with rheumatoid arthritis in their control group. This difference might be explained by the fact that these studies used different control groups. However, Blenkiron et al.[104] also used a healthy control group and in contrast with White and Schweitzer, they found the values for perfectionism on the MPS to be *lower* in their CFS sample than in their healthy control group. This example brings us to another issue in the possible explanation of the lack of uniformity in the major findings of the studies.

Another possible reason for a lack of consistency in the major findings could be that not all studies used comparable control groups. Whereas many studies used healthy individuals as (part of) their control group[94,97-100,102,105-107], others used patients with fibromyalgia/ chronic pain[96,107], depressed patients[100,101,106], patients with multiple sclerosis[93,102,106], or patients with rheumatoid arthritis[103]. As a consequence, the results of the studies can only be interpreted relative to the specific control groups that were used. Certain differences between patients with CFS and controls that might be obvious with one control group, might become less significant, or even get completely lost with another.

So, the results of a specific study can only be interpreted in the light of the control group that was used. However, this is of course rather common in medical and psychological research. Be that as it may, in the case of CFS the same applies to the patient groups that were included, which is far less usual. While most studies used adult patients with CFS, one study used adolescent patients with CFS of whom most were recovered[98] and another study exclusively included combat exposed Gulf War veterans with CFS[99]. Nevertheless, this would seem to leave all the studies using 'ordinary' adult individuals with CFS to be comparable. However, as different CFS case definitions were used, this is not the case. Some studies used the original CDC criteria of 1988[3], others the revised CDC criteria of 1992[95], others the revised CDC criteria of 1994[8], others the UK operational criteria of 1991[7] and one New Zealand's McKenzie criteria of 1988[108]. To add to the confusion and making the different findings even more difficult to compare, some studies distinguished between noninfectious and postinfectious CFS patients[94], some studies distinguished between patients with CFS and co-morbid psychiatric

disorder/ depression and patients with CFS without co-morbid psychiatric disorder/ depression[99,100], and one study only included non-depressed patients with CFS[101]. This brings us to the next important problem, the influence of depression on the study of CFS and personality.

Several studies on the psychiatric status of patients with CFS were discussed in the introduction. However, as was mentioned there, depression is not an exclusionary criterion for the diagnosis of CFS and therefore inevitably plays an important role in the personality studies on CFS. As was noted by Buchwald[111] and Wessely et al.[112], amongst others, there is a considerable overlap between the criteria used for several psychiatric DSM-diagnoses (most notably depression) and CFS. As a consequence patients with symptoms required for a diagnosis of CFS, at the same time have symptoms fitting into a diagnosis of depression.

When distinguishing between patients with or without depression, some found that depression had a great impact on the major findings of their study. In the study by Fiedler et al.[99], the CFS with psychiatric co-morbidity group scored significantly higher than the CFS without psychiatric co-morbidity group on the neuroticism subscales of anxiety, hostility, self-consciousness, impulsivity and vulnerability. Chubb et al.[100] found that the scores of patients with CFS were not different from those of healthy controls, except for those subjects with CFS who were concurrently depressed, where the scores resembled the scores of their depressed control group. Johnson et al.[106] also found that most of the personality disorders in their CFS group were accounted for by the CFS group with concurrent depressive disorder. However, in contrast with these findings, Henderson and Tannock[92] concluded that they were *unable* to account for the presence of personality disorder in their assessment of patients with CFS, by comorbid depression. An additional problem is that the Beck Depression Inventory (BDI), which three of the studies used[103,106,107], was found to perform poorly as a screener for depression in subjects with CFS[113].

All in all, the role of depression in CFS is extremely difficult to determine as there are at least three plausible relationships. It could be that depression is a predisposing, causative factor in CFS. On the other hand, it might be that *"CFS is no more than depression masquerading as a physical illness"* (p. 2)[5], but it is also possible that depression is a reaction to the illness and to the lack of clarity that surrounds CFS. In this case it would be likely that depression is caused by the stress of being diagnosed with a disease of unknown origin, in combination with the absence of a standard treatment and the possible disbelief encountered in the health care setting. As it seems to be the case with many of the findings of abnormalities in CFS, the role of depression in the pathogenesis and perpetuation of CFS remains unclear. These questions of causality and nosology however, are somewhat beyond the

reach of this article and will therefore not be discussed further**. Nevertheless, by raising these questions we get to a more fundamental level of inquiry. In the next section some conceptual issues regarding the study of personality in CFS will be addressed.

Conceptual background of personality studies in CFS

As mentioned, the methods used to study personality in CFS are quite diverse. Nonetheless, in the approach of the reviewed studies, a shared conceptual model regarding the possible association of personality and CFS, and the appropriate way to scientifically study it, seems to be reflected.

Firstly, these studies have focused much of their attention on personality *disorder*. Psychological *malfunctioning*, rather than ordinary, non-pathological and everyday aspects of personality, which are commonly seen as a primary concern of personality psychology, has been a main interest of personality research in CFS so far. By such a focus on, and an overrepresentation of the psychopathological aspects of personality, it is easy to provide only a one-sided and too stringent image of the personality of individuals with CFS.

Secondly, on the whole these studies have tended to conceptualize personality mainly in its most general and decontextualized structures. With the use of psychological tests like the Tridimensional Personality Questionnaire, the NEO Five-Factor Inventory and the Eysenk Personality Questionnaire, certain characteristics of personality, such as extroversion, neuroticism and social desirability can accurately be studied and compared. However, in this way personality is approached primarily in its most basic and undifferentiated structure, and only a limited understanding of personality is provided[119]. Although personality traits can provide a kind of dispositional signature of the person, few links have been made between traits and actual contextualized behavior[120] and it seems unlikely that the exclusive knowledge of such a basic structure of relatively nonconditional and noncontingent dispositional traits, or psychopathological personality profiles, is enough to *wholly* explain and account for the behavioral consequences of CFS, or the complex association between personality and the syndrome.

Within the humanities and the social sciences, especially personality psychology, there has been an increasing awareness that persons do not *merely* act and experience on the basis of quantifiable, general traits. They primarily evaluate and motivate their behavior and beliefs in qualitative, contextualized terms[121,122]. On the basis of these terms, persons assess their behavior, interpret themselves, articulate what they believe to be important, try to make sense of their past, give meaning to the present, direct their future projects and provide their life with purpose and unity[123]. Personality is not a static, independent, self-

contained and decontextualized 'given', but is always dynamically constructed in dialogue with others, and against a 'meaningful' background provided by social practices and culturally shared moral values[124,125]. In recent decades, the idea of the 'narrative' has emerged as a new metaphor not only within personality psychology[126,127], but also within clinical psychology[128-130]. From this approach, persons are understood as the creators of meaning, and narrative thought is seen as the process by which these meanings are developed and changed[131]. The narrative is seen not only as a novel way of conceptualizing human experience and identity, but also as an useful clinical tool to help individuals understand why they act, and organize their lives, in certain ways, and to aid them in retelling and reorganizing their lifestory. In a broader concept of personality, than that which was used so far in the research on CFS, the lifestory could be seen as a special kind of psychosocial construction and individuals might be understood as trying to coauthor a thematically coherent and meaningful narrative with, and against the background of, their culture and social world.

Dispositional traits and life narratives can be regarded as two different levels of personality[132], each with their own methods of study, frameworks and taxonomies. In CFS, personality traits are usually studied through the use of standardized questionnaires and (semi) structured interviews in the search for abnormalities, or deviations from the average. The benefit and attraction of studying personality in this way is not only that it is rather time and cost effective, but also that it produces objective, quantifiable and comparable data and as such seems to be in accordance with the rigorous methods of the natural sciences. The downside to this approach is that, to a considerable extent, it decontextualizes human experience and behavior from its real life setting, and its social and cultural background. The usefulness and attraction of studying personality on the level of the life narrative, on the other hand, is that it can remain much closer to the continually evolving and subjectively experienced reality of the person. Starting from the assumption of normality, personality on this level is usually studied through an open dialogue in which the subject matters are decided, not primarily by the investigator, but in the first place by the person him- or herself. Just as with personality considered on a trait level however, the benefits to this approach also entail its main drawbacks. Besides being rather time-consuming, the obtained data might be difficult to compare and, because of their specific temporal and spatial context, be of a contingent and subjective nature. This can lead to the assumption that personality, considered as a developing lifestory changing through time, cannot be categorized, quantified or systematically researched[123] and that it, because of this, cannot be studied in a proper methodical way.

Within the scientific debates on CFS, some have tried to draw attention to the fact that the lifestories of the patients seem to have been neglected. Van

Houdenhove[133] for example, states that *"much of the etiological and therapeutic controversies about the so called chronic fatigue syndrome (CFS)[…] may be due to the relative neglect of the patient's story – in clinical practice as well as in research. More specifically I believe that insufficient attention is being paid to the mostly significant context in which the illness began, and the possible connection between the illness and the patient's life history. [T]he patient's biography should be part of each diagnostic evaluation and considered an important focus of psychological/ psychiatric research in CFS"* (p. 495). At present, there have been few who have addressed these concerns. Some qualitative studies have described, in narrative terms, the experience of patients of the impact of CFS as a disruption and disorganization of their premorbid lifestory and identity. The transformation and rewriting of those stories is depicted as an inescapable consequence of getting CFS and is usually followed by a subsequent quest for the restoration and reorganization of a meaningful autobiographical self-narrative[134-136]. Currently however, the biggest challenge for those wishing to systematically study the association of personality, considered on the narrative level, and CFS, will be to do so with methods that are firmly based in psychological theories about personality and psychotherapy and that have been specifically designed to analyze and categorize a person's narrative into its most meaningful temporal constituents. Moreover, such methods should be psychometrically validated and not only allow a study of the idiosyncrasies of the single case, but these methods must also have been developed in such a way that they can be generalized to a population and that quantitative comparisons between different groups can be made[137-139].

CONCLUSION

Every science, whether it be psychology, medicine, physics or sociology, is based on a set of conceptual assumptions. Usually, when these disciplines are functioning satisfactory, these presuppositions remain implicit and there is no need to make them explicit. However, when problems arise that seem difficult to solve with the normal instruments of these sciences, we have to focus our attention explicitly on these conceptual assumptions and ask ourselves whether our understanding of the problem is not somehow obscured by the commonly accepted model of thought. For psychology and medicine, CFS poses exactly such a problem.

In this article the first aim was to give a concise review of the current research on personality and CFS. There seems to be consistent evidence that patients with CFS often score higher on some personality traits, most notably neuroticism, than healthy controls. Furthermore, higher levels of DSM axis II diagnoses, most notably obsessive-compulsive, histrionic and borderline personality disorders, within the CFS population, in comparison to healthy populations are found. *However*, there are some important confounding elements in these findings. When compared to patients with another chronic

illness, the finding of specific personality differences is far less common and usually annulled. Additionally, the finding of divergence could often be explained by co-morbid depression/ psychiatric disorder. Another limitation is that, at times, instruments have been used to study certain aspects of personality (e.g. the MMPI, the BDI and the MCMI) that have later been found to perform inadequately for patients with CFS. And lastly, many studies eventually conclude that the found personality differences are consequences of the disease, rather than precipating factors and as such play no causal role in CFS. All in all, under careful scrutiny the previously mentioned stereotype of patients with CFS does not seem to be justified. Nonetheless, at present there do seem to be at least three *overarching* conclusions that can be drawn with regard to personality and CFS. Firstly, the heterogeneity of findings *within* the CFS groups implies that, on the trait or psychopathological level, there are no unique personality characteristics that are either a necessary condition for, or an unavoidable consequence of CFS. Secondly, although personality traits such as neuroticism and perfectionism are generally considered to be stable, non-conditional and not effected by life changes[140], most studies seem to agree on the possibility that the pre-morbid personalities of their subjects might have changed as a result of their condition. Diverse forms of chronic illness seem to be able to alter personality in similar ways and increased levels of neuroticism and introversion for example (not to mention depression), could well be a feature of many different diseases. In fact, the American Psychiatric Association acknowledges the possibility of personality change as a result of chronic illness[141]. Thirdly, as a consequence of this, it can be concluded that cross-sectional designs in the long run will probably not be able to provide definitive answers to the question of the exact role of personality in CFS.

In the section on the methodological problems of these personality studies it was suggested that *some* of the confusion that remains regarding the association between personality and CFS might be due to a variety in study methods, control groups and CFS case definitions. This diversity seems almost unavoidable. However, with regard to control groups, the substantial overlap between CFS and some psychiatric diagnoses (e.g. depression), and other unexplained medical conditions (e.g. fibromyalgia) is truly confusing in research, and makes patients from these populations difficult to compare. Age and sex matched healthy individuals, and patients with a somatic illness in which fatigue is also a main complaint (e.g. rheumatoid arthritis, multiple sclerosis) seem to be much better suited as control groups. With regard to study methods, the exclusion of general psychopathology or shared problems on a dispositional trait level in CFS has of course been essential and valuable in the personality research on CFS. However, the cross-sectional designs of the reviewed studies make inferences about causality very difficult. New insights might be gained by longitudinal designs, studying the predictive validity of

certain personality traits as risk factors for the development of CFS. Prospective studies in clinical populations of mood disorders and emotional risk factors in relation to CFS for example, have already been able to provide some evidence regarding their precipating role[142,143]. In addition, it will prove insightful to follow-up a cohort of patients with a relatively short illness duration (i.e. a recent diagnosis of CFS) in order to study whether certain personality characteristics, and levels of depression, change as a consequence of prolonged illness duration.

Finally, in the section on the conceptual background of the study on personality in CFS, it was argued that although the methods used so far were diverse, the studies seemed to share some basic conceptual assumptions regarding personality and the way to study it. Up to now, personality research in CFS has either been in search for personality disorder and psychological malfunctioning, or has been conducted on a general, non-relational trait level. Nevertheless, the fact that to a large extent personality is something that can only exist in, and develop through the inherent relations and dialogues with family, peers, colleagues, media, society and culture in general, must be taken in account. A similar perspective has been brought forward[144,145] with regard to the understanding of the personality and (mental) health within collective cultures, but it also seems particularly true in the study of CFS. Future personality research in CFS should not only take the abovementioned methodological issues into account and be of a more longitudinal nature, but should also be directed towards, and become aware of the dialogically constructed, historically contextualized and indissoluble relational terms by which persons understand, evaluate and articulate themselves.

Modern individualized society, to a considerable degree, is focused on achievement, consumption and success, and is characterized by a plurality of rapid economical, political, religious, technological and cultural changes. Against this background, modernity confronts people in a whole new fashion with a multiplicity of problems and possible ways of life and the need, and imperative, to find and develop a meaningful identity. New insights into the possible difficulties and stumbling-blocks in the personality of individuals with CFS might be gained, if research attention would also concentrate on systematically and comparatively studying individuals with CFS, as socioculturally embedded agents who are trying to construct a coherent and intelligible self-narrative.

FOOTNOTES

* Millon et al.[91] being the first to study the role of personality in CFS, was included although the study lacked an appropriate control group.

** See, for example, Abbey and Garfinkel[114], Moss-Morris and Petrie[115], and Swartz[116] for some of the articles concerned with the relation between CFS and depression, Wessely et al.[2] and Aaron and Buchwald[1] for a more general discussion concerning the nosological status of CFS, and Bolton[117] and Borch-Jacobsen[118] for a more philosophical, historical discussion of nosological problems in the definition of psychiatric disorders.

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Chapter 2

Adolescents' self-positioning in chronic fatigue syndrome & juvenile idiopathic arthritis

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Submitted

ABSTRACT

Objective: In dialogical self theory, the self is regarded as incorporating multiple positions, or perspectives. Physical and psychosocial impairment, related to chronic illness, might alter the organization of these self-positions. In this study, the self-positioning in adolescent chronic fatigue syndrome (CFS) and juvenile idiopathic arthritis (JIA) will be investigated. **Method:** 42 adolescents with CFS, 37 adolescents with JIA and 23 healthy teenagers completed a standardized version of the Personal Position Repertoire procedure (PPR) -a quantitative method to analyze the organization of self-positions-, the Child Health Questionnaire (CHQ-CF87) -assessing physical and psychosocial functioning-, and the Checklist Individual Strength (CIS-20) -measuring fatigue. **Results:** Adolescents with CFS reported severe physical difficulties and psychosocial problems. They positioned themselves as significantly less strong and more unwell in their social relationships in comparison to healthy adolescents and patients with JIA. While adolescents with JIA reported bodily pain, low general health and impaired physical functioning, they positioned themselves very similar to healthy teenagers. In their social contexts, they presented themselves mainly as strong and healthy. **Conclusions:** In this study, the focus was on the everyday manifestation of adolescents' selves in their social contexts. Especially for adolescents with CFS, there is a strong indication that positions relating to illness are (overly) dominant in their self-organization. Teenagers with JIA show a contrasting, strong self-positioning approach. While this might be adequate and sustainable in adolescence, it could prove too strenuous to maintain throughout adult life. It is regarded of clinical importance to address these issues in this crucial developmental period.

INTRODUCTION

A major challenge of adolescence is to attain a sense of self that is coherent, unique, and continuous over time[1]. In the transition from childhood to adult life, identity starts to mature and a new balance between autonomy and communion is sought. During adolescence, an exploration of a plurality of self-identities is common, socially accepted and probably even necessary[2]. Prolonged sickness can substantially hinder this development[3]. Although this aspect of adolescent mental health is of clinical importance, as a well-balanced identity can support successful coping and adjustment to long-lasting illness[4], it is not much studied in pediatric settings.

This article focuses on adolescents with (*a*) the chronic fatigue syndrome (CFS) –a medically unexplained condition, mainly characterized by persistent and severe fatigue[5], and (*b*) juvenile idiopathic arthritis (JIA) -a rheumatic disease mainly characterized by unpredictable and repeated episodes of joint inflammation and pain[6]. In previous (adult) research on CFS in relation to self-identity, patients with the syndrome have expressed the feeling of no longer having control over their own lives[7]. They describe a lack of physical and social activities, and a deprivation of the various emotions these activities entail, leading to a feeling that opportunities for true engagement with the world have been cut off[8,9]. Furthermore, patients indicate that certain impoverished and unwanted positions of the self have gained a dominant role, seemingly impervious to change[10,11]. In the relatively few studies on the relation between self-identity and arthritis, patients with arthritis have described their search for an autonomous identity, their wish to achieve a meaningful and normal life, and their desire to participate fully in society[12,13]. In addition, they report a loss of social roles, and they express experiences of stigmatization and discrimination, leading to feelings of decreased self-worth[14,15].

These studies provide insight into self-identity in CFS and arthritis. Nonetheless, at present all research in this field has been qualitative. In addition, most investigations have been conducted without control groups. This makes it difficult to interpret the findings as relative to a specific condition, and to distinguish the outcomes from the general consequences of prolonged illness. Furthermore, very few studies have focused specifically on adolescence as a crucial period in identity formation. Especially in this developmental phase however, identity is not only an intra-individual, but also very much an interpersonal process[16,17].

Dialogical self theory is a relatively new current in identity research, incorporating quantitative methodology, and sensitive to the contextual dimensions of individuals' self-formation[18]. This theory starts from the notion that identity emerges from a plurality of positions[19]. The dialogical self is understood, not as one single and continuous 'I', but instead as a dynamic

multiplicity of self-positions[20,21]. Trying to structure and balance a multiplicity of positions is seen as one of the challenges of self-identity in adolescence[22].

At present in clinical research, dialogical self theory and methodology have mainly been used to study and conceptualize problems of identity in adult psychiatric disorder[23-25]. It has not yet been employed in the research into adolescent pediatric conditions. In this study, dialogical self theory and methodology was employed to gain more insight into possible identity issues in adolescent chronic illness. It was hypothesized that physical and psychosocial impairment, related to chronic illness, might alter the organization of self-positions. Therefore, the self-positioning, the level of fatigue, and the physical and psychosocial functioning of healthy adolescents was compared to that of adolescents with CFS, and adolescents with JIA.

METHOD

Participants

The total group consisted of healthy adolescents (*n* =23), adolescents with JIA (*n* = 37), and adolescents with CFS (*n* = 42). In table 1, their general characteristics are presented. The higher percentage of female participants in this study is in accordance with the general predominance of women in both (adolescent) CFS, and JIA[26,27]. All participating adolescents were invited by their pediatrician at the Department of Pediatrics (*n* =79), or in two local high-schools (*n* = 23) to volunteer in this study. Study criteria dictated that *a*) each participant was between 14 and 19 years old, *b*) did not have a primary psychiatric or psychological diagnosis, *c*) the patients were diagnosed with CFS[28], or JIA[29] by a pediatrician, and *d*) the healthy adolescents did not have a medical condition. The study was approved by the ethical committee of the university medical center, and informed consent was obtained from all participating adolescents and their parents, or caregivers.

Personal Position Repertoire procedure (PPR)

In dialogical self theory, a distinction is made between internal and external positions. Internal positions are regarded as those positions, that are relevant to the particular ways in which individual people organize their lives (e.g. I as adaptive, I as strong, I as unwell). From the perspective of these internal positions, those people in the social environment that are relevant, are referred to as external positions (e.g. my father, my teacher). The PPR is a method to investigate basic aspects of the relations between internal and external positions[30]. In the PPR, internal and external positions are combined into a matrix, in which the internal positions form the rows, and the external positions form the columns. In completing such a matrix, individuals are asked to estimate on a 6-point Likert scale (ranging from 0 = not at all, to 5 = in a very strong degree), their answer to the question *"In which degree is this internal*

position (I as…) *prominent in the contact with this external position* (My…)*?"*. This is done for all internal positions, in relation to all external positions.

A standardized list of a variety of internal positions was used, to assess a broad array of positions, important in the self-identity of (chronically ill) adolescents. Each adolescent individually formulated the most important external positions in his or her life. To enable the use of this information in statistical analysis, the external positions were divided into five categories relevant for adolescents; 1) direct family (i.e. parents and siblings), 2) indirect family (e.g. grandparents, aunts, or nephews), 3) school (e.g. teachers, or classmates), 4) friends, and 5) adversaries.

Physical functioning, psychosocial functioning and fatigue measurements

Physical and psychosocial functioning was assessed with the Child Health Questionnaire-Child Form (CHQ-CF87). The CHQ-CF87 is a reliable, validated assessment measure, with good internal consistency (average Cronbach's α of 0.80)[31]. Fatigue was assessed with the subjective fatigue subscale of the Checklist Individual Strength (CIS-20). This scale is a reliable, validated assessment measure, with good internal consistency (Cronbach's α of 0.93)[32].

Design and Statistical Analysis

With hierarchical cluster analysis (Ward's method, Squared Euclidean distances on standardized scores), and principal components analysis, the internal positions were divided into discrete subgroups in order to enable group comparisons. The PPR-data were analyzed with SPSS-GLM (general linear model) as a GROUP x DOMAIN "split plot design". In order to analyze the data within a split plot design, the internal PPR-clusters were aggregated across five domains for every person. Global main and interaction effects were tested for. Subsequently, the differences in internal positioning, between the three groups, were analyzed. Data were now aggregated for every person, and simple one-way ANOVA's were performed, using post-hoc Bonferroni comparisons. All statistical analysis was performed using SPSS, version 15.0.

Table 1
General Characteristics

	CFS (*n* = 42)	JIA (*n* = 37)	Healthy (*n* = 23)	*p*
Age				
mean in years (SD)	16.4 (1.1)	16.3 (1.5)	16.4 (0.8)	.92
Gender (% girls)	83	84	78	.84
Ethnicity *n* (%)				
Caucasian	42 (100)	33 (89)	20 (88)	
African	-	3 (8)	1 (4)	
Asian	-	-	1 (4)	
Latin-American	-	1 (3)	1 (4)	
Disease duration	1.6 (1.0-3.5)	7.5 (3.5-12.4)	-	.00
median in years (IQR)				
Type of JIA, n (%)				
systemic[a]	-	3 (8)	-	
polyarticular[b]	-	25 (68)	-	
oligoarticular[c]	-	9 (24)	-	

Note. [a] Most severe form of arthritis with fever, rash, and inflammation of other organs; [b] Affecting ≥ 5 joints; [c] Affecting < 5 joints.

RESULTS

Internal Positioning in Relation to External Positions

The hierarchical cluster analysis resulted in 6 clusters of internal positions:

Cluster 1, *'strong'* (Cronbach's α = .86) consisted of the internal positions: 'I as strong', 'I as healthy', 'I as energetic', 'I as self-confident', 'I as decisive', 'I as active', 'I as optimistic', and 'I as satisfied'.

Cluster 2, *'adaptive'* (Cronbach's α = .63) consisted of the internal positions: 'I as adaptive', 'I as perfectionist', and 'I as caring'.

Cluster 3, *'vulnerable'* (Cronbach's α = .75) consisted of the internal positions: 'I as vulnerable', 'I as anxious', and 'I as doubtful'.

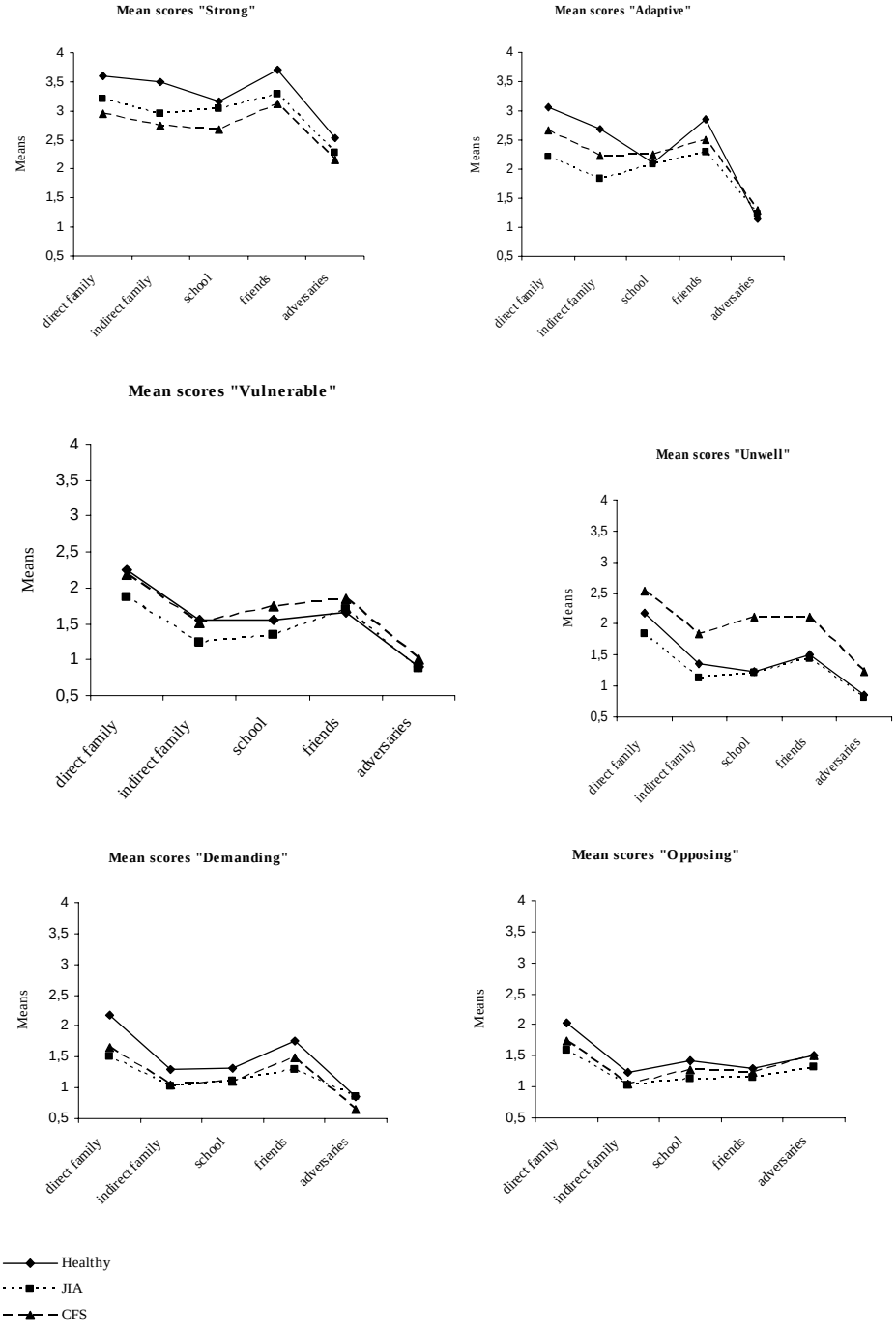
Cluster 4, *'unwell'* (Cronbach's α = .81) consisted of the internal positions: 'I as unwell', 'I as tired', 'I as victim', and 'I as dependent'.

Cluster 5, *'demanding'* (Cronbach's α = .66) consisted of the internal positions: 'I as demanding', 'I attention-seeking', and 'I as jealous'.

Cluster 6, *'opposing'* (Cronbach's α = .70) consisted of the internal positions: 'I as quarrelsome', 'I as dissatisfied', 'I as pessimistic', 'I as unmotivated', 'I as lazy', and 'I as avoiding'.

In figure 1, the mean-scores of the internal positions for the three groups of adolescents in relation to their external positions are presented. Overall, only one significant interaction was found, namely on the cluster 'adaptive' (Wilk's

Figure 1
Mean scores for the six clusters on five categories of external positions.



lambda = .79, F (8,186) = 2.85, p =.005). As can be seen in the six graphs, all three groups show a similar pattern for most of the clusters of internal positions. In other words, for all of the adolescents, whether healthy or ill, the same internal positions are prominent in the contact with the same group of external positions. The mean scores of direct family and friends in contrast to indirect family, school and adversaries, had significant estimated contrasts (two-sided p-value <.001) for all of the six clusters ranging from ψ = .236 to ψ = .729; indicating that internal positioning is context dependent.

Overall Internal Positioning

In table 2, the mean overall score on the six clusters of internal positions are presented. The adolescents with CFS showed their *'strong'* internal positions significantly less than the healthy adolescents, and they showed their *'unwell'* internal positions significantly more. Furthermore, the adolescents with CFS also showed their *'unwell'* positions significantly more than the adolescents with JIA. Adolescents with JIA only differed from their healthy peers in that they showed their *'adaptive'* internal positions significantly less (p = .031).

Table 2

Means *(M)*, standard deviations *(SD)* and F-values *(F)* for internal positions

	CFS	**JIA**	**Healthy**	
Clusters	***M (SD)***	***M (SD)***	***M (SD)***	***F***
Strong	2.8 (0.8)[a]	3.1 (0.8)	3.4 (0.4)[b]	6.55**
Adaptive	2.3 (0.8)	2.0 (0.8)[a]	2.6 (0.8)[b]	3.56*
Vulnerable	1.8 (0.8)	1.5 (0.9)	1.7 (0.5)	0.79
Unwell	2.1 (0.6)[a]	1.4 (0.8)[b]	1.5 (0.7)[b]	9.56**
Demanding	1.3 (0.8)	1.2 (0.8)	1.6 (0.7)	1.74
Opposing	1.4 (0.7)	1.7 (0.8)	1.5 (0.7)	0.90

Note. * p < .05. ** p < .005. Means with different superscripts differ significantly at p < .05.

Physical functioning, psychosocial functioning and fatigue

In table 3, the physical, psychosocial functioning and fatigue scores are presented. Adolescents with CFS and JIA scored significantly lower than healthy teenagers on physical functioning, bodily pain and general health perception. In addition, adolescents with CFS also scored significantly lower than healthy teenagers and adolescents with JIA on self-esteem, mental health, and physical role functioning.

Table 3
Fatigue, physical and psychosocial functioning

	CFS	JIA	Healthy	*F*
Physical functioning (9 items; 0-100)†	68.7 (17.0)[a]	77.0 (25.4)[a]	96.4 (4.7)[b]	16.6**
Bodily pain (2 items; 0-100)†	39.0 (23.7)[a]	50.6 (25.2)[a]	69.6 (20.5)[b]	12.9**
General health perceptions (13 items; 0-100)†	38.8 (18.9)[a]	57.0 (16.7)[b]	76.3 (10.4)[c]	40.3**
Role functioning: Emotional (3 items; 0-100)†	65.9 (33.2)	80.9 (26.8)	82.9 (18.2)	3.9
Role functioning: Behavioral (3 items; 0-100)†	83.9 (26.8)	92.9 (19.2)	89.8 (18.2)	1.6
Role functioning: Physical (3 items; 0-100)†	36.8 (23.7)[a]	81.8 (26.3)[b]	84.3 (22.0)[b]	44.4**
General behavior(16 items; 0-100)†	74.1 (13.4)[a]	82.1 (10.6)[b]	80.3 (11.0)	4.8**
Mental health (16 items; 0-100)†	56.9 (14.1)[a]	71.8 (14.5)[b]	70.7 (9.9)[b]	14.4**
Self esteem (14 items; 0-100)†	57.4 (12.6)[a]	68.8 (11.7)[b]	71.7 (6.6)[b]	16.0**
Subjective fatigue (8 items; 8-56)‡	48.1 (7.0)[a]	29.0 (13.2)[b]	26.7 (10.8)[b]	46.7**

Note. †Higher scores indicate better physical or psychosocial well being. ‡High score indicates a high level of subjective fatigue. * $p < .05$. ** $p < .001$. Means with different superscripts differ significantly at $p < .05$.

DISCUSSION

In this study, adolescents' self-positioning in their social contexts was examined. Obvious similarities were found between the groups, in relation to their social positioning. Although in varying degrees, for adolescents with or without a chronic disease the same internal positions are prominent in their different social contexts. Family and friends are confronted with the broadest array of internal positions. In these contacts, adolescents can freely experiment with different internal positions, resulting in situations in which opposing, as well as strong positions can alternate with unwell and vulnerable positions. With other external positions on the contrary, the relationship is typically characterized by a more selective positioning.

Healthy adolescents, not dealing with the challenges caused by a chronic illness, prominently show their strong and adaptive positions. Adolescents with CFS on the other hand show their strong positions significantly less than their healthy peers. Furthermore, they show their unwell internal positions not only significantly more than healthy teenagers, but also than adolescents with JIA. Severe fatigue (whatever its cause) will likely entail a profound alteration of, and a withdrawal from adolescents' usual experiences. In the prolonged (sensed) absence of other self-positions to occupy and stories to tell, one position (or group of positions), 'I as unwell' for example, could become increasingly dominant in the adolescents' position repertoires. Stronger or more optimistic internal positions could become limited, leading to a self-perpetuating cycle of reduced physical and social activity. Furthermore, despite vigorous research into its possible causes, so far no etiology for (adolescent) CFS has been found, and it remains a rather controversial diagnosis. This ambiguity of the status of CFS as a medical condition, and the experienced stigmatization by others, might also influence the way in which the positioning repertoires of the adolescents with CFS are organized. To overcome this perceived stigma, the patients might be inclined to focus on their unwell internal positions, in order to convince their surroundings of the severe and real impediments of their condition.

The self-positioning of adolescents with JIA shows a striking resemblance to that of the healthy adolescents, although the mean scores are (non-significantly) lower for most clusters. In general, they appear to cope relatively well with the consequences of their painful and potentially incapacitating illness. For family, clinicians and researchers however, it is often difficult to believe that the possible weight gain and transformed appearance, the joint inflammation, and physical limitations in JIA, would not have social and behavioral consequences. In fact, in the research literature on the outcome of JIA, it is pointed out that, in young adulthood, individuals who have (had) arthritis show higher unemployment rates and reduced physical health, more psychosocial impairment, and a lower quality of life[33,34]. This 'transitioning problem' might be (partially) due to a self-positioning approach that, while adequate and sustainable in adolescence, could prove to be too strenuous to maintain throughout adult life. As stated by Stanton, Revenson and Tennen[35], *"unbalanced attention to positive adjustment to chronic illness can also have untoward consequences. The expectation of the unfailingly 'strong' patient permits the ill person little latitude for having a bad day (or a bad year)"* (p. 568). During adolescence, the mainly positive and strong position repertoires of these youngsters can possibly be maintained because of social support in this life-phase; protective parents, an accommodating school with its continuity and safety, encouraging peer relations, and a pediatric hospital in which personal attention is central. In this situation (in which the environment seems to mostly adapt itself to them)

adolescents with JIA might have less need to adopt their '*adaptive'* positions. However, transition to adulthood may entail critical challenges for these adolescents, due to inherent changes in their living circumstances; finding a job, living on one's own, committing to new relationships, and the transition to adult medical care, in which a protective approach is less obvious.

The adolescents with CFS, as well as the adolescents with JIA differed significantly from their healthy peers on bodily pain, general health perception and physical functioning. In everyday life however, it seems as though especially the patients with CFS are limited by this physical impairment, as can be witnessed in their low physical role functioning scores. Additionally, in comparison to the healthy teenagers, these patients also had significantly worse scores on measurements of fatigue, mental health, and self-esteem. One could argue that it is especially these more psychosocial components of health status that are related to identity problems. However, due to the cross-sectional design of this study definite conclusions with regard to this relation cannot be drawn. Therefore, follow-up studies investigating longitudinal changes in adolescents' identity processes in relation to their health status are of principal importance. It should also be noted that self-positioning is just one aspect of the dialogical self, and that important notions such as narrative and affect where not directly investigated in the present study. Moreover, it must be mentioned that while the participating adolescents completed a standardized version of the PPR, in a clinical setting it is probably more beneficial to have clients freely indicate and chose those internal positions that they recognize, and identify with. Furthermore, although there is evidence that psychological difficulties are not related to disease activity[36], it is imaginable that the quite severe illness symptoms and prolonged illness duration in our patient groups – related to the tertiary, academic hospital setting- could have had an effect on our findings. It should be acknowledged that these factors were not specifically taken into account. Future research might also want to focus on less severely affected patients, or patients at an earlier stage of their illness.

CONCLUSION

This dialogical self theory-based study has been the first to quantitatively investigate adolescent CFS and JIA in relation to identity conceptualized on the level of self-positioning. While most psychological studies into adolescent CFS and JIA have focused on psychosocial dysfunctioning and disorder, in this study we concentrated primarily on the everyday expression of adolescents' identity in their social contexts. We studied whether CFS and JIA would lead to an altered organization of adolescents' positioning repertoires. There is a strong indication that physical and especially psychosocial impairment in adolescence, and the identity level of self-positioning are related. These findings therefore have important implications for clinical practice as well. The

organization of self-positions is not only something that can be studied by researchers, it can also be scrutinized by the patients themselves in self-investigation[37]. For adolescent patients with CFS and JIA alike, it might be very helpful to investigate their different self-positions, to try to include and develop new positions, to form coalitions between existing positions, and to balance previously dominant positions with counter-positions in the experiences of everyday life. In this way communications between self-positions can be recovered, and new feelings, possibilities and perceptions (both positive and negative) can be opened[38].

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Chapter 3

Adolescents' emotional experience in chronic fatigue syndrome & juvenile idiopathic arthritis

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Submitted

ABSTRACT

Objective: To study the emotional experience of adolescent patients. **Method:** The emotional experience of 36 adolescents with juvenile idiopathic arthritis (JIA), 42 adolescents with chronic fatigue syndrome (CFS), and 25 healthy teenagers was studied with the Self-confrontation Method (SCM). The SCM is used to analyze the affective level of personal narratives. With the Child Health Questionnaire (CHQ), health-related quality of life was assessed. **Results:** Adolescent patients reported less positive experiences of autonomy and success, compared to healthy adolescents. Furthermore, adolescents with CFS described more negative experiences of powerlessness and isolation. In the CHQ-results, both patient groups scored lower on physical scales than healthy peers. Adolescents with CFS also scored lower on mental health, self-esteem, and physical role-functioning. **Conclusions:** While the presence of negative experiences plays a role in the CFS group, the absence of certain positive experiences is apparent in both adolescent patient groups. These findings can sharpen therapeutic focus.

INTRODUCTION

There is widespread recognition that chronic illness does not only have consequences on a biological level, but also on a psychological and a social level[1]. These consequences may profoundly alter emotional experiences, influencing the general functioning and well-being of patients. The affective level should therefore be considered as an important target for therapeutic intervention[2,3]. For example, in a recent study investigating the 'active' ingredients of cognitive behavior therapy for chronic fatigue (as a medically unexplained symptom), it was found that emotional processing, including the expression, acknowledgement and acceptance of emotional distress, was the key predictor of a good outcome[4].

Most research on emotion and health has a focus on the negative impact of "negative" emotions, supporting the view that these negative emotions advance illness, whereas positive emotions promote health. Although emotional problems have usually been considered to reflect the presence of negative affective states, and the amelioration of negative emotions has traditionally been a focus of psychotherapy, emotional experience might also become problematic when certain positive affective states are absent[5,6]. Research on quality of life in arthritis, for example, has shown that patients with higher levels of pain and activity limitation reported less adaptive coping, associated with lower positive affect[7].

Adolescence is a period characterized by multiple changes. Challenges on a bodily, personal and social level, have to be integrated in identity formation. In the case of chronic illness, factors like chronic pain, severe prolonged fatigue, or uncertainties about improvement, have to be incorporated in a coherent sense of self. In integrating a variety of (positive as well as negative) emotions into research on therapy[8], there appears to be a lack of reliable, widely applicable and valid methods of eliciting, and measuring emotional experience on the level of implicit meaning[9,10]. It has been suggested that these techniques, measuring emotional experience, should involve the exploration of personal narratives in relation to their affective associations[11,12]. Gaining insight in the organization of personal narratives of adolescent patients might be a step in the direction of broadening our understanding of the various patterns of emotional responses in (and to) chronic illness.

In this study, we applied the self-confrontation method (SCM) as an instrument to structurally assess and analyze personal narratives on an explicit level, along with the intrinsically related emotional experience on an implicit meaning level[13]. This study used the SCM to compare the personal narratives and emotional experience of adolescents with chronic fatigue syndrome (CFS) -a condition of unknown etiology, mainly characterized by seriously disabling fatigue[14], and adolescents with juvenile idiopathic arthritis (JIA) –an oftentimes severely impairing, chronic auto-immune disorder, to a large extent

characterized by unpredictable episodes of joint inflammation[15]. While at present there have been many investigations into cognitive and behavioral factors involved in CFS and JIA[16,17], there are relatively few studies into the emotional experience of adolescent patients with these conditions. The objective of the present study was to examine the emotional experience, positive and negative, of adolescents with CFS and JIA. We expected these groups to differ from healthy adolescents, not only on the manifest level of personal meaning, but also particularly on the latent level of emotional experience. We hypothesized that a) experiences with negative emotions are more prominent in the life stories of adolescents with CFS and JIA than in those of their healthy peers, and that b) experiences with positive emotions are less prominent in the life stories of adolescents with CFS and JIA than in those of their healthy peers.

METHOD

Participants

The participants included healthy adolescents (n =25), adolescents with JIA (n = 36), and adolescents with CFS (n = 42). Their general characteristics are presented in table 1. The percentage of female participants in this study is in accordance with the general predominance of women in both CFS and JIA[18,19]. All participating adolescents were asked by their pediatrician in the University Medical Center (n =78), or in two local high-schools (n = 25), to volunteer in this study. Study criteria dictated that a) each participant was between 14 and 19 years old, b) did not have a primary psychiatric or psychological diagnosis, c) the patients were diagnosed with CFS[20] or JIA[21] by a pediatrician, and d) the healthy adolescents did not have a medical condition. The study was approved by the ethical committee of the University Medical Center. Informed consent was obtained from all participating adolescents and their parents.

Table 1
General Characteristics

	Healthy (*n* = 25)	JIA (*n* = 36)	CFS (*n* = 42)
Age			
mean in years (*SD*)	16.5 (0.8)	16.3 (1.5)	16.4 (1.1)
Gender (% girls)	80	83	83
Ethnicity *n* (%)			
Caucasian	20 (80)	32 (89)	42 (100)
African	1 (4)	3 (8)	-
Asian	3 (12)	-	-
Latin-American	1 (4)	1 (3)	-
Disease duration	-	7.7 (3.5-12.6)	1.6 (1.0-3.5)
median in years (IQR)			
Type of JIA, *n* (%)			
systemic[a]	-	3 (8)	-
polyarticular[b]	-	24 (67)	-
oligoarticular[c]	-	9 (25)	-

Note. [a] Most severe form of arthritis with fever, rash, and inflammation of other organs; [b] Affecting ≥ 5 joints; [c] Affecting < 5 joints.

SCM-Procedure

The SCM is a well-established, evidence-based psychological assessment measure[22] and clinical tool, developed to study individual experiences and their ordering into a meaning system through self-investigation - with special attention to affective organization. It's theoretical background[23], its clinical use[24,25], its psychometric properties, validity and reliability[26,27], and its use to compare groups[28], have all been extensively described.

As a start of the SCM-procedure, the researcher presents a set of open questions in order to elicit relevant parts of the adolescent's life story. Subsequently, the adolescent formulates the most important units of meaning from the past, present and future into written sentences. A self-investigation with adolescents generally consists of 20 to 35 sentences. These sentences are explicit and contain beliefs, images, wishes and relationships. The underlying narrative theme and emotions however, are implicit and can be more difficult to reveal. Therefore, after the formulation of these sentences, a standard list of 24 affect terms is presented. These affects are subdivided into four scales that are universally found in psychological literature, and that are psychometrically sound and well suited for use in research and practice[29]:

1) *P*ositive (P): *joy, happiness, enjoyment, trust, security, and inner calm* (Cronbach's α = .93).

2) *N*egative (N): *powerlessness, anxiety, shame, self-alienation, guilt, loneliness, inferiority, and anger* (Cronbach's α = .90).

3) *S*elf (S), reflecting 'the striving for self-enhancement': *self-confidence, strength, self-esteem, pride, energy, and freedom* (Cronbach's α = .91).

4) *O*ther (O), reflecting 'the longing for contact and union with the other': *caring, love, tenderness, and intimacy* (Cronbach's α = .86).

For each particular sentence, the adolescent is asked to indicate on a 6 point Likert scale (0 = not at all, and 5 = very much) to what extent each affect is experienced. Furthermore, in order to get insight into the present, overall situation, the adolescent is asked to use the 24 affect terms to evaluate a response to the question *"How do you generally feel these days?",* and *"How would you like to feel?"* In addition, the adolescents are asked to characterize the experience of their disease by affectively rating the sentence: *"My CFS…"*, *"My JIA…"*, or *"When I'm ill…",* respectively for the adolescents with CFS, JIA, and the healthy adolescents.

Sentences of meaningful experiences with a great variety on the manifest level, may show similarities on the affective level. By computing product-moment correlations between the affective profiles of every sentence of the adolescent, similarities on the implicit level of the personal narrative can be found. Such correlations do not intend to specify a causal relation, but rather an affective similarity. In table 2, the sentences that have the highest product-moment correlation with the standardized sentence concerning disease (D, in table 2), general feeling (G, in table 2) and ideal feeling (I, in table 2) are given, formulated by three adolescents from the CFS group, and three adolescents from the JIA group.

Table 2
Sentences correlating with the standardized sentences and scores on S, O, P and N

Sentences of adolescents with CFS	**S**[a]	**O**[a]	**P**[a]	**N**[a]	***r***
CFS 1:					
D) My headaches in elementary school were partly caused by problems I had in my class.	0.8	0.8	0.6	1.4	.64
G) My brother had an appendicitis operation with complications and shortly after that he pulled a pot of boiling oil over himself.	0.8	2.0	0.8	1.9	.68
I) I was surprised that I got a good grade for my language test.	4.0	0	2.0	0.3	.60
CFS 2:					
D) I often feel afraid and I have the feeling that people can't help me with that. This feels like a part of me that I can't change.	1.3	1.0	0.5	3.5	.95
G) My boyfriend has a large influence on my life. We fight and quarrel all the time.	2.3	3.8	1.4	3.1	.70
I) My dream is to have a job and earn a lot of money, have a nice house with my partner and to do lots of fun stuff. I want	4.8	4.3	4.6	0	.94

kids and pets.					
CFS 3:					
D) To my father, my brother was holy and I always got the blame for everything.	0	0	0	3.3	.94
G) 4 years ago my fatigue, pain and concentration problems started. From that time on everything that happened to me is a blur.	0	0	0	3.3	.93
I) Three months ago I started riding a horse again.	4.0	4.0	4.3	0.3	.95
Sentences of adolescents with JIA					
JIA 1:					
D) When I think of my past, I remember that I always had to be aware of my arthritis. When other kids did a physical activity, I could not participate.	0.8	0.8	0.1	1.6	.83
G) When I'm 18, I will visit Colombia with my twin brother.	1.8	1.0	2.4	0.1	.78
I) I am an example for many of my peers, both for good as well as bad things. They believe me and listen to me.	3.0	.75	2.9	0	.86
JIA 2:					
D) I would rather not think about my future and arthritis.	1.5	0	0.4	1.5	.86
G) I get along very well with my brother and we talk a lot.	3.3	1.3	3.8	0.1	.84
I) In three years I see myself in a nursing education, because working with mentally disabled people seems impressive and important to me.	4.5	3.3	4.6	0	.88
JIA 3:					
D) Especially if I cannot do certain things due to my arthritis, I really want to do them.	1.5	0	0.9	1.3	.74
G) In the last 4 years I have gotten a very close group of friends with whom I do everything together.	4.5	4.3	4.8	0.1	.96
I) During the holidays I had a fantastic time with all my friends.	5.0	4.3	4.6	0.1	.97

Note: S = mean score of affect reflecting self-enhancement, O = mean score of affect reflecting desire for contact with others, P = mean score of positive affect, N = mean score of negative affect, r = product-moment correlation with standard sentence: D) sentence with the highest correlation with disease related standard sentence, G) sentence with the highest correlation with the general feeling, I) sentence with the highest correlation with the ideal feeling, [a] Six-point Likert scale 0-5, Cronbach's alpha reliability coefficients for the subscales were 0.86-0.93.

In order to find the central theme in teenagers' personal narratives, the sentence representing how the adolescent generally feels is used as pivotal sentence. Concentrating on the shared meaning between all sentences with a strong positive correlation ($r > .60$) with the sentence concerning the general feeling, the adolescent is asked to review this central theme of his or her personal narrative. To find possible contrasts in the personal narrative, the

final step of the SCM-procedure is to search for sentences with the highest *negative* correlation ($r > -.60$) with the most generalizing sentence. In table 3, as an illustration, the themes of the same adolescents as in table 2 are presented. In this way, the thematic organization of the system is elicited, thereby offering insight into the differentiation of the total system of experiences, and revealing the opposing sides of the central theme of the adolescent's personal narrative[30].

SCM-typology

From the combination of sentences with their affective organization, general types of experiences can be discerned. The six most common, psychometrically validated types of experiences[29] are summarized based on a combination of the four different affect dimensions (Positive, Negative, Self, and Other): *'Strength and Union'*, *'Aggression and Anger'*, *'Autonomy and Success'*, *'Unfulfilled longing'*, *'Unity and Love'*, and *'Powerlessness and Isolation'*.

Physical and psychosocial functioning

The Child Health Questionnaire-Child Form (CHQ-CF87) was used to assess the physical and psychosocial functioning of patients and healthy adolescents[31]. The CHQ-CF87 is an 87-item generic self-administered instrument which measures physical, emotional, and social components of health status in children and adolescents, independently of the underlying disease. Cronbach's alpha reliability coefficients for the subscales of the CHQ were 0.69-0.92.

Statistical Analyses

The data were analyzed with one-way ANOVA's, focusing on three 'a priori contrasts': JIA vs. Healthy, CFS vs. Healthy, and CFS vs. JIA. In preparation for these comparisons several measures were aggregated from the original SCM-matrices (n = 103). The aggregation of the information of the sentences consisted of two steps. First, every sentence was classified according to the typology, using the S-O-P-N profile. Subsequently, the percentages of the six types of experiences were aggregated for every person. All statistical analyses were performed using SPSS, version 16.0.

Table 3
Themes of the six adolescents resulting from the correlation with the general feeling

Themes of adolescents with CFS:
CFS 1:
+) When I do things to attain my *own* goals, or when I do things that I initially dread doing, I feel satisfied, proud and happy.
-)* Problems that I experience in tense, new, or unreasonable situations make me feel anxious, powerless and angry. I try to forget or suppress these feelings.
CFS 2:
+) If I do what *I* really want, I feel at best. **-)*** I'm stuck in a situation in which I feel powerless, frightened and alone. I don't know what to do and I don't think other people can help me. There is a lot going on in my head and that's why I find it hard to relax, I can't sleep well and I can't concentrate. This makes my fatigue worse.
CFS 3:
+) I feel like I am myself, energetic and proud, when I'm active, when I'm doing something enjoyable, or when I have a nice contact with someone. Then I can let go of everything, I have distraction and everything goes naturally. **-)*** If I have the feeling that I don't have matters in my own hand (the way people judge me, when I feel aversion doing something, or if I can't be myself), I get a strong negative feeling (less self-assured, inferior, angry and frightened), and that takes away a lot of my energy.
Themes of adolescents with JIA:
JIA 1:
+)* When I feel connected with others, I can be myself and show my willpower. In such situations I feel free and happy. **-)** I always have to be careful with my arthritis, especially with physical activities. I think about it a lot, but I sometimes treat the subject too lightly.
JIA 2:
+)* I feel good and self-assured if I can talk to someone about myself, and when I do fun things with people I like. Then I can look at the present and future from a positive perspective. **-)** Negative, annoying things I don't discuss with others. I'm afraid they will find me pathetic and a poseur. This makes me feel powerless and lonely, so I would rather avoid and suppress these feelings.
JIA 3:
+)* I care a lot about the people I love. They make it easy to focus on the good things, instead of the bad. So I am actually quite dependent on them, because I always need one of them around. **-)** If I feel dependent because of my arthritis I always persist. I have a hard time asking people for favors in such situations and feel like I'm a pain in the ass, or pathetic.

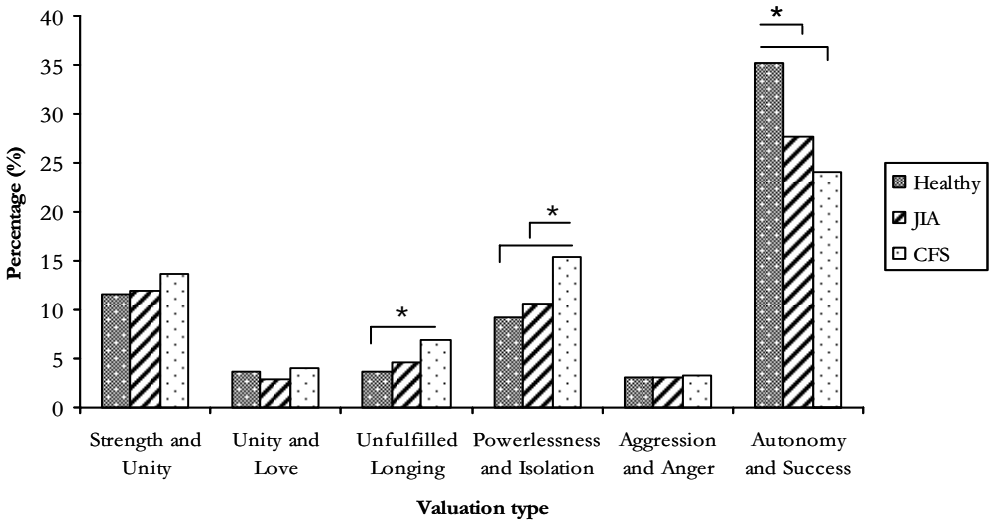
Note. +) positive side of the theme, -) negative side of the theme, * the highest correlation with the general feeling

RESULTS

Typology

In total, the 103 adolescents formulated 2424 sentences (M = 23.5, SD = 4.7) concerning their past, present and future. With a priori contrasts, a comparison was made of the proportion of the different types of experiences within the total set of sentences, for each adolescent of the three groups. Significant differences were found for the proportion of experiences of *'Autonomy and Success'*, '*Unfulfilled longing'*, and '*Powerlessness and Isolation'* (figure 1). Healthy adolescents formulated an average of 35.2 % of experiences of *'Autonomy and Success'*. This percentage was significantly higher than the proportion of these experiences for adolescents with JIA (27.6 %, t (100) = 1.697, $p < .05$), and for adolescents with CFS (24.1 %, t (100) = 2,558, $p < .001$). In addition, adolescents with CFS had a higher proportion of experiences of *'Powerlessness and Isolation'* (15.4%) than their healthy peers (9.2 %, t (100) = 2.003, $p < .05$); and also than the adolescents with JIA (10.5 %, t (100) = 1.761, $p < .05$), who didn't differ from their healthy peers in the further distribution of the types of experiences. Finally, adolescents with CFS had a significantly higher proportion of experiences of '*Unfulfilled longing'* than the healthy adolescents (6.9 % respectively 3.7 %, t (100) = 1.639, $p = .05$).

Figure 1.
Proportion (%) of types of experiences within adolescents' total narrative systems



Note. * $p < .05$.

Physical and psychosocial functioning

In table 4, the physical and psychosocial functioning scores are presented. Both adolescents with CFS and adolescents with JIA scored significantly lower than the healthy controls on 'physical functioning', 'bodily pain', and 'general health perceptions'. The remaining scores for the health concepts of adolescents with JIA were comparable to the mean values of the healthy subjects. Adolescents with CFS showed worse values on *all* health concepts compared to both adolescents with JIA and healthy adolescents. These differences in physical and psychosocial functioning were statistically significant for the following scales: 'mental health', 'self-esteem', and 'role functioning: physical'.

Table 4
Physical and psychosocial functioning

	CFS	JIA	Healthy	*F*
Physical functioning (9 items; 0-100)	68.7 (17.0)[a]	76.5 (25.7)[a]	96.4 (4.8)[b]	16.0**
Role functioning: Emotional (3 items; 0-100)	65.9 (33.2)	80.6 (27.1)	82.1 (18.3)	3.6
Role functioning: Behavioral (3 items; 0-100)	83.9 (26.8)	92.7 (19.4)	89.4 (18.5)	1.5
Role functioning: Physical (3 items; 0-100)	36.8 (23.7)[a]	81.3 (26.5)[b]	83.6 (22.2)[b]	42.2**
Bodily pain (2 items; 0-100)	39.0 (23.7)[a]	51.1 (25.6)[a]	68.3 (19.9)[b]	11.4**
General behavior (16 items; 0-100)	74.1 (13.4)[a]	82.5 (10.5)[b]	79.5 (10.6)	5.0*
Mental health (16 items; 0-100)	56.9 (14.1)[a]	72.6 (14.0)[b]	70.4 (10.0)[b]	15.4**
Self esteem (14 items; 0-100)	57.4 (12.6)[a]	68.8 (11.9)[b]	71.7 (6.8)[b]	15.5**
General health perceptions (13 items; 0-100)	38.8 (18.9)[a]	57.6 (16.6)[b]	76.3 (10.6)[c]	39.4**

Note. Items scored on a Likert scale; higher scores indicate better physical or psychosocial well being. * $p < .05$. ** $p < .001$. Means with different superscripts differ significantly at $p < .05$.

DISCUSSION

Each personal narrative is unique, on a manifest as well as on an emotional, latent level. This makes narratives notoriously difficult to analyze and compare quantitatively. In this study, the focus was on the search for similarities and differences on the emotional level, between three groups of adolescents. The emotional experience of all adolescents showed apparent similarities, especially with regard to emotional experiences referring to 'strength and unity', 'aggression and anger', and 'unity and love'. In the personal narratives of healthy adolescents more than one third of the experiences referred to 'autonomy and success'. The most obvious difference between healthy teenagers and patients is that the personal narratives of adolescents dealing with the challenges involved in CFS or JIA, contained a significant smaller proportion of these experiences of 'autonomy and success'. Integrating this specific type of experience into a personal narrative is generally needed for identity formation, one of the main developmental tasks of adolescence.

Chronic illness, especially in adolescence, can limit the possibilities to develop strong feelings of self-enhancement as it can increase dependence on parents due to physical limitations (e.g. parents bringing their adolescent child to school, or a party with friends), cause a more negative body image (e.g. JIA can cause deformations, lack of physical activities can result in weight gain), and may decrease confidence in their personal future (e.g. insecurity of disease progress, choice for a profession must be adapted to physical abilities).

Furthermore, contrary to our expectation, adolescents with JIA did not report significantly more experiences of a negative affect type. Compared to adolescents with JIA and their healthy peers, adolescents with CFS reported a significantly higher proportion of experiences referring to 'powerlessness and isolation' in their personal narratives. This is often related to situations in which the individual feels that there is no way out, and feelings of numbness, hopelessness and helplessness can be related to this type of experiences. In addition, compared to their healthy peers, adolescents with CFS had significantly more experiences of 'unfulfilled longing'. This type of experience is directed to the adoption or maintenance of a loving orientation toward another person (or object) that is, or seems, unreachable.

Scoring the CHQ, adolescents with CFS, as well as adolescents with JIA differed significantly from their healthy peers on physical functioning, bodily pain, and general health perception. In everyday life however, it seems as though especially the patients with CFS are limited by this physical impairment, as can be witnessed in their low physical role functioning scores, and their significantly worse scores on mental health, and self-esteem. This might be reflected in their SCM-results by their higher proportion of negative experiences in their personal narratives

It should be noted, that the SCM requires the adolescents to have a basic capacity for self-reflection, which is related to a certain developmental age, cognitive, and emotional level of functioning. Furthermore, the quite severe illness symptoms and prolonged illness duration of our patient groups – related to the tertiary, academic hospital setting- could have had an effect on our findings. These factors, along with disease activity, were not specifically taken into account. In this study, there was a considerable variability in duration of illness. Adolescents with JIA were seen later in the course of their illness than adolescents with CFS. It is difficult to predict how this might have influenced our results. Patients with a longer period of illness might experience less emotional distress with time. In contrast, adolescents further from the time of diagnosis who experience ongoing symptoms might exhibit greater levels of emotional distress. Furthermore, CFS and JIA have quite a different nosological status. Whereas JIA is considered a clearly somatic (although idiopathic) condition, CFS is regarded as a functional somatic syndrome[32]. Patients with CFS often report to feel stigmatized by others who tend to doubt

the status of CFS as a medical condition[33]. The potential influence of illness duration and stigmatization should be studied in future research.

CONCLUSION

With our findings in mind, we suggest that psychological intervention should focus on the integration and utilization of *positive,* as well as negative emotions[34,35]. Direct reflection upon emotional experience, and the immediate alteration of the affective level should be considered an important focus of psychological treatment as it is closely associated with beneficial therapeutic change[36,37]. These considerations are reflected in, what we believe to be, the most important clinical implication of the present investigation. While the presence of negative emotions certainly seems to play a significant role in our CFS group, the absence of certain positive emotions appears to be of importance in the emotional experience of both our JIA group, as well as our CFS group. By eliciting the personal narratives of adolescent patients, and subsequently relating these to experienced emotions, a direct insight into the underlying thematic organization of these patients' life stories, could be gained. More important: this is an insight that the adolescents gain *themselves* through self-investigation. Information that can be gained through standardized questionnaires (like the CHQ) is now made explicit by, and to the patient him- or herself, in a personalized way. In a therapeutic setting, as well as in daily life, the self-discovered narrative theme can serve as a starting point from which adolescents can explore in what concrete personal situations, their theme is manifested. Following such an assessment, adolescents can experiment (under therapeutic supervision) with the creation of new personal meanings and the fortification of new themes[30]. Rather than focusing on a general negative topic (e.g. fatigue or pain), this insight into an adolescent's unique theme, in relation to its actual manifestations on an emotional and experiential level, allows for a much more specific employment of various therapeutic interventions and techniques, adjusted to the adolescents' own perspective.

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Chapter 4

Adolescent chronic fatigue syndrome: A follow-up study

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ABSTRACT

Objective: To describe the symptomatic and educational long-term outcome, healthcare use, and risk factors for non-recovery in adolescent chronic fatigue syndrome (CFS). **Method:** A follow-up study of 60 adolescents with CFS after regular care. The Checklist Individual Strength (CIS-20) and the Child Health Questionnaire (CHQ-CF87). General questionnaire regarding further symptoms, school attendance, work attendance and treatment. **Results:** Complete measurements for 54 adolescents were returned (90%). At initial assessment, their mean age was 16.0 years (SD, 1.5), and 20.4% was male. Mean follow-up duration was 2.2 years. At follow-up, the mean age was 18.2 (SD, 1.5), 28 (51.9%) adolescents had a near to complete improvement, but 26 (48.1%) were not improved. Teenagers who attended school (n, 41) had missed an average of 33% of classes during the last month. The rest (n, 13) had worked an average of 38.7% of a fulltime job during the last month. 66.7% of subjects were treated by a physiotherapist, 38.9% were clinically treated in rehabilitation, 48.1% had received psychological support, and 53.7% had used alternative treatment. **Conclusions:** About half of the adolescents had recovered from CFS at follow-up. The other half was still severely fatigued and physically impaired. Health care use had been high, and school- and work attendance was low. Older age at inclusion was a risk factor, and pain, poor mental health, self-esteem, and general health perception at outcome, were associated with an unfavorable outcome. Future research should focus on customizing existing treatment and studying additional treatment options.

INTRODUCTION

The chronic fatigue syndrome (CFS) in adolescence is a heterogeneous and medically unexplained condition[1]. No laboratory tests for (adolescent) CFS are available[2]. Its main symptom is functionally disabling fatigue, severely affecting (young) patients' lives [3]. Like in adult CFS, the most commonly used criteria in adolescent CFS are the English Oxford[4], and the US Centers for Disease Control and Prevention (CDC)[5] criteria. The Oxford criteria are generally considered somewhat less restrictive[6]. The prevalence of adolescent CFS has been estimated at 1.3-4.4% in British and US populations[7,8]. Estimations of the prevalence of adolescent CFS in other populations are sometimes lower[9]. The incidence of adolescent CFS is estimated at 0.5%[10], and the female to male ratio is estimated at 4:1[11]. Randomized controlled treatment trials for adolescent CFS have been rare, but there is growing support for a positive effect of cognitive behavior therapy[12,13].

There have only been a few follow-up studies describing the outcome of adolescent CFS after regular care, applying either Oxford, or CDC-criteria for CFS. In table 1, the main studies on the prognosis of adolescent CFS are presented. About one-third, to a half of the patients in the presented studies still experienced severe fatigue, physical impairment and little improvement at follow-up. At present, the largest cohort described in a follow-up study of pediatric CFS included 35 patients. Most studies so far have used the Oxford criteria for CFS, and there is only one follow-up study that has employed the CDC (1994) criteria for CFS. However, all diagnoses in these studies were made retrospectively. Furthermore, most studies had a disproportionately high percentage of male participants and a wide age range. In this study, the outcome of 54 adolescents, fulfilling the CDC (1994) criteria for CFS is described.

Table 1
Follow-up studies of adolescent CFS

Study	n[a]	Age[b]	Gender[c]	Duration[d]	Criteria[e]	Outcome
Smith et al. (1991)[14]	15* (100%)	14.5 (1.7)	40%	1.1-2.7	CDC (1988)†	26.7% recovered from illness; 26.7% marked improvement; 46.7% unimproved or worse.
Rangel et al. (2000)[16]	25 (78.1%)	11.7 (2.2)	40%	3.8	Oxford†	36% recovered; 32% mildly fatigued and impaired; 32% unimproved or worse.
Bell et al. (2001)[17]	35** (76%)	12.1 (5-18)	31.4%	13.0	Patients diagnosed before 1988†	37.1% recovered from illness; 42.9% better, but not resolved; 11.4% chronically ill; 8.6% more ill.
Gill et al. (2004)[18]	16 (72%)	14.9 (2.4)	25%	4.1	CDC†	25% near to complete improvement; 31% partial improvement; 44% unimproved or worse.
Sankey et al. (2006)[20]	28 (60%)	13.3 (7-17)	54%	3.0	Oxford†	66.7% improved; 33.3% unimproved or worse.

Note. Table might not include all research on the outcome in adolescent CFS (according to the Oxford, or the CDC-criteria), but to the authors' best knowledge represents the main studies in the field. [a]Number of participants (response rate, %). [b]Age at onset in years (SD; otherwise range). [c]Percentage of male participants (%). [d]Duration of follow-up; mean in years (SD; otherwise range). [e]Diagnostic criteria used in study. *5 subjects were also identified with major depression (including some who were also diagnosed with CFS), the respondent was either the adolescent or the adolescent's parent, and eventually only six patients fulfilled CDC (1988) criteria for CFS **Patients were part of a cluster outbreak of CFS, and authors' own CFS-criteria were used[18]. †Diagnosis established retrospectively.

METHOD

Participants

All participating adolescents had first visited a general practitioner before being referred to a general pediatrician in a non-academic setting. Subsequently, they were referred to the academic pediatric hospital. 74 adolescents who had previously participated in a number of research studies into adolescent CFS[21,22] were considered for inclusion. At initial examination, these adolescents were assessed for CFS and the diagnosis, in accordance with the CDC-1994 criteria, was either made or confirmed by a specialized academic pediatrician. Although they had been clinically diagnosed with CFS, 14 of the 74 adolescents (18.9%)

were not eligible for participation in this follow-up study because their initial scores on the subjective fatigue subscale of the Checklist Individual Strength (CIS-20) were below cut-off (see next section). Questionnaires were sent to the remaining 60 adolescents. All questionnaires were filled out at home. The duration of the follow-up was defined as the time between the initial research examinations at the academic pediatric hospital, and the present study's assessments. The study was approved by the ethical committee of the hospital, and informed consent was obtained from all participating adolescents and their parents.

Primary outcome measures

Fatigue was assessed with the subjective fatigue subscale of the CIS-20[23]. This scale measures experienced fatigue, and consists of 8 items; scores range from 8 (no fatigue) to 56 (extremely fatigued). It is a reliable, validated assessment measure, with good internal consistency (Cronbach's α 0.93[12]). The CIS-20 has previously been used in research into adolescent CFS[22]. Various cut-off scores (ranging from 35.7-40) for recovery on this measure have been employed[24,12]. In this study the cut-off on this subscale was set at 40 (mean, plus 2 SD of the subjective fatigue distribution in healthy adolescents[24]), to dichotomize outcome as improved (<40) or not-improved (≥40).

Functional impairment was measured with the physical role functioning subscale of the Child Health Questionnaire-Child Form (CHQ-CF87)[25]. This scale measures limitations in school work and daily activities as a result of physical health, and consists of 3 items; scores range from 0 (severe limitations, due to physical problems) to 100 (no limitations, due to physical problems). It is a reliable, validated assessment measure, with good internal consistency (Cronbach's α 0.86[26]). The CHQ-CF87 has previously been used in research into adolescent CFS[23]. Cut-off scores for recovery on the physical role functioning subscale of the CHQ-CF87 have not yet been set for adolescent CFS. However, on the physical functioning subscale of the Short-Form General Health Survey (SF-36)[27], also ranging from 0 (maximal physical limitation) to 100 (ability to do all activities) and previously employed in adolescent CFS, a cut-off of 65 has been used[12]. In this study, the cut-off on the physical role functioning subscale of the CHQ-CF87 was correspondingly set at 65, to dichotomize outcome as improved (>65) or not-improved (≤65). A classification of near to complete improvement required a score <40 on the subjective fatigue subscale of the CIS-20, combined with a score >65 on the physical role functioning subscale of the CHQ-CF87.

Secondary outcome measures

In addition to the physical role functioning subscale of the CHQ-CF87, we employed the emotional role functioning subscale, measuring limitations in

school work and daily activities as a result of emotional problems, such as worry or sorrow (Cronbach's α 0.90[26]), the behavioral role functioning subscale, measuring limitations in school work and daily activities as a result of behavioral problems (Cronbach's α 0.71[26]), the bodily pain subscale, measuring severity and frequency of bodily pain (Cronbach's α 0.85[26]), the general behavior subscale, measuring the exhibition of aggressive, delinquent and immature behavior (Cronbach's α 0.79[26]), the mental health subscale, measuring a diversity of positive and negative feelings (Cronbach's α 0.86[26]), the self-esteem subscale, measuring satisfaction with abilities, looks, family/ peer relations and life overall (Cronbach's α 0.89[26]), and the general health perceptions subscale, measuring believes concerning health (Cronbach's α 0.77[26]), in order to cover additional physical and psychosocial domains at outcome.

In a further general questionnaire, the participants were asked to indicate (yes/ no) the regular presence during the last month of 8 symptoms in accordance to the CDC-1994 criteria (self-reported impairment in memory or concentration, sore throat, tender cervical or lymph nodes, muscle pain, multi-joint pain, headache, unrefreshing sleep, post-exertional malaise lasting 24h or more). School attendance was measured as the percentage of classes the adolescent had attended school during the last month, compared to the school schedule of class-mates. If the participants no longer attended school, work attendance was calculated as the percentage of a fulltime job (38h) the adolescent had worked during the last month. Finally, participants were asked to estimate the total number of therapeutic visits they had (if any) with physiotherapists, psychologists and alternative healthcare suppliers in the follow-up period, and (if applicable) to estimate the months they had spend in clinical rehabilitation.

Analyses

All statistical analyses were performed using SPSS (version 16.0). On the outcome variables, group means and standard deviations were calculated. Potential risk factors (e.g. gender, age, severity of fatigue at inclusion, duration of follow-up) were quantified through odds ratios using logistic regression with outcome (recovered/ not recovered) as dependent variable. The significance level was set at $p<.05$ (two-tailed tests).

RESULTS

Complete data for 54 (90%) adolescents were returned. At first inclusion, their mean age was 16.0 years (SD, 1.5), all were Caucasian, 20.4% was male, and their mean score on the subjective fatigue subscale of the CIS-20 was 49.4 (SD, 5.1). The onset had been gradual in 27 (50%) of the cases, following 'flu-like illness' in 22 (40.7%) of the cases, and 'acute' (i.e. of a sudden onset, without

preceding 'flu-like symptoms') in 5 (9.3%) of the cases. At follow-up, the mean age was 18.2 years (SD, 1.5). There were no significant differences between responders and non-responders in gender, age or fatigue severity. The mean follow-up duration was 2.2 years (SD, 1.6), but the symptoms in most cases had existed substantially in the years prior to initial assessment. During this time, 43 (79.6%) adolescents had not received any other diagnosis, 3 (5.6%) were diagnosed with celiac disease, 3 (5.6%) with lactose intolerance, 2 (3.7%) with metabolic disorder, 1 (1.9%) with hypermobility syndrome, 1 (1.9%) with major depressive disorder, and 1 (1.9%) with anxiety disorder.

Table 2 shows the scores on the primary outcome measures of the 54 adolescents for whom complete measurements were returned. At follow-up, 28 adolescents (51.9%) had a score <40 on the subjective fatigue subscale of the CIS-20, and a score >65 on the physical role functioning subscale of the CHQ-CF87, indicating that they had a near to complete improvement from CFS at follow-up.

Table 2
Primary outcome measures

	Mean (SD)	**Number of patients below (CIS-20), or above (CHQ-CF87) cut-off n (%)**
Checklist Individual Strength (CIS-20)[a]		
Subjective fatigue	34.3 (14.1)	30 (55.6%)*
Child Health Questionnaire (CHQ-CF87)[b]		
Role functioning: Physical (3 items; 0-100)	71.9 (28.3)	38 (70.4%)**
Near to complete improvement, n (%) [c]	---	28 (51.9%)***

Note. [a]Score ranges from 8-56; a high score indicates a high level of fatigue. [b]Score ranges from 0-100; a high score indicates better physical functioning. [c]Primary outcome measure. *Score <40 **Score ≥65 ***Subjective fatigue subscale (CIS-20) <40, and physical role functioning subscale (CHQ-87) >65.

Table 3
Secondary outcome measures

	Mean (SD)	Number of patients above (CHQ-CF87), or below (CDC-criteria) cut-off n (%)
Child Health Questionnaire (CHQ-CF87)[a]		
Role functioning: Emotional (3 items; 0-100)	84.9 (25.2)	46 (85.1%)*
Role functioning: Behavioral (3 items; 0-100)	95.8 (10.5)	52 (96.3%)*
Bodily pain (2 items; 0-100)	56.1 (27.9)	22 (40.7%)*
General behavior(16 items; 0-100)	82.0 (10.7)	49 (90.7%)*
Mental health (16 items; 0-100)	68.8 (14.9)	34 (63.0%)*
Self esteem (14 items; 0-100)	66.2 (14.6)	28 (51.9%)*
General health perceptions (13 items; 0-100)	46.8 (18.7)	10 (18.5%)*
CDC-criteria[b]		
Additional symptoms	3.8 (2.3)	20 (37%)**

Note. [a]Scores range from 0-100; higher scores indicate more well-being. [b]Score ranges from 0-8; a high score indicates more symptoms in accordance with CDC-1994 criteria. *Score ≥65 **Score <4

Table 3 shows the scores on the secondary outcome measures. The mean scores on the behavioral role functioning, the emotional role functioning, and the general behavior subscales of the CHQ-CF87 were generally good. However, the mean scores on the mental health and self esteem subscales of the CHQ-CF87 were unfavorable (approximately 1 SD below the mean scores in a healthy young population[26]), and especially the mean scores on the bodily pain and general health perceptions subscales of the CHQ-CF87 were low (respectively about 1.5 and 2 SD below the mean scores in a healthy young population[26]).

Table 4 shows the school attendance, work attendance and therapeutic contacts of the 54 adolescents for whom complete measurements were returned. At follow-up, the participants who still attended school had on average missed approximately one-third of the regular classes during the last month. For a previously described cohort of 167 healthy adolescents this percentage was only 12.5%[21]. In The Netherlands it is common to start a fulltime job after finishing school. However, those participants who no longer attended school had worked only an average of approximately one-third of a fulltime job during the last month. The variety and frequency of therapeutic healthcare use had been considerable between initial assessment and follow-up.

Only a higher age at initial inclusion was found to be a risk factor for non-recovery of CFS at follow-up (odds ratio, 1.59; 95% CI, 1.06-2.39; *P*=.03). Gender, severity of fatigue at inclusion, type of onset, other diagnoses, health

care use (psychological treatment, physiotherapy, rehabilitation or alternative treatment), and length of time between inclusion and follow-up were not associated with outcome. At follow-up, a high amount of reported CDC CFS-symptoms (odds ratio, 1.62; 95% CI, 1.19-2.22; *P*=0.00), and a low score on the mental health (odds ratio, 0.95; 95% CI, 0.90-0.99; *P*=.01), self esteem (odds ratio, 0.94; 95% CI, 0.90-0.99; *P*=.01), bodily pain (odds ratio, 0.93; 95%CI, 0.93-0.99; *P*=.00), and general health perceptions (odds ratio, 0.92; 95% CI, 0.84-0.96; *P*=0.00) subscales of the CHQ-CF87 were associated with non-recovery.

Table 4
School attendance, work attendance and health care use at follow-up

School attendance and work	
Adolescents attending school, n (%)	41 (75.9%)
Percentage of classes followed, % (SD)	67.0% (34.2)
Adolescents working*, n (%)	13 (24.1%)
Percentage of full-time job worked**, % (SD)	38.7% (35.0)
Health care use	
Physiotherapy, n (%)	36 (66.7%)
Mean number of contacts (SD)	45.9 (46.6)
Alternative treatment, n (%)	29 (53.7%)
Mean number of contacts (SD)	16.8 (15.5)
Psychological support***, n (%)	26 (48.1%)
Mean number of contacts (SD)	32.6 (32.6)
Clinical treatment in rehabilitation, n (%)	21 (38.9%)
Mean months in treatment (SD)	3.6 (2.2)

Note. *Only those adolescents no longer attending school. **38 hours per week. ***General non-CFS specific CBT was common in routine psychological support

DISCUSSION

The outcome of unexplained pediatric chronic fatigue (i.e. not diagnosed as CFS) is mostly positive[28,29]. It is generally thought that the prognosis for adolescent CFS is also relatively good[1,2]. In this study it was found that, although about half of the participating adolescents had a near to complete improvement, the other half was still severely fatigued, had impaired physical functioning, and would probably still fulfill CDC-1994 criteria for CFS at follow-up.

The cohort of adolescents participating in this study is the largest described in any follow-up after regular care. The diagnosis of CFS was established according to the CDC-1994 criteria at initial examination, the

female to male ratio was in accordance with research findings, the mean age of participants did not have a wide range, and a strict cut-off score, on a validated measure for adolescent CFS, was used to qualify subjects for inclusion. However, the CHQ-CF87 was not employed at inclusion. Therefore, potential predictors for outcome were limited. Furthermore, while all questionnaires were validated for this age group and none of the adolescents indicated that they had difficulty with completion, the answering of questionnaires at home might not ensure complete confidentiality without parental influence.

In previous studies, few risk factors for (adolescent) CFS have been identified[30]. Like in our study, an older age at diagnosis has been found to imply an increased risk for prolonged adolescent CFS[3,10]. The high levels of school non-attendance are in concordance with the literature[31,1,2]. The use of health care services in adolescent CFS was high and also comparable to recent studies[32,33]. While no specific form of received health care was associated with a better outcome, almost all adolescents had received some kind of treatment and it is difficult to estimate what the outcome would have been had this not been the case. Although some of the participating adolescents had received CBT as part of routine psychological support, this also did not result in a superior outcome. This is consistent with findings that the results of CBT for CFS are generally superior within, rather than outside the confines of randomized controlled trials[34]. The percentage of adolescents who had not recovered from CFS at follow-up in this study was somewhat higher than in the previous studies and more like the outcome rates in adult CFS[35]. This might be due to the strict use of CDC-1994 criteria for CFS, the employment of a cut-off score for eligibility, and a higher age of participants at inclusion. In addition, the diagnosis of CFS in the adolescents participating in this study was established (or confirmed) in a tertiary, academic hospital setting and might represent a particularly impaired cohort.

CONCLUSION

Despite intensive health care use a substantial proportion of adolescent patients with CFS remain severely fatigued and physically impaired. This is associated with considerable bodily pain, poor mental health, self esteem and general health, and impacts greatly on school- and work attendance. Therefore, future research into adolescent CFS should not only focus on recognizing patient characteristics for a favorable outcome, but should also be directed towards further customizing existing treatment, and studying additional interventions for those patients that do not benefit from established treatment options[36,37].

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Chapter 5

Self-investigation in adolescent chronic fatigue syndrome: Narrative changes and health improvement

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ABSTRACT

Objective: A small-scale intervention study into narrative self-investigation in adolescent chronic fatigue syndrome (CFS). **Method:** The Self-confrontation Method (SCM) is an instrument to assess and change personal life stories. Forty-two adolescents diagnosed with CFS were included and randomly assigned to either 6 or 12 sessions with the SCM. Twenty-five healthy adolescents were assigned to 6 sessions. Outcome was measured directly after the self-investigation procedure at 4 months. Follow-up measurements were made 10 months later. The Checklist Individual Strength and the Child Health Questionnaire were used to measure changes in fatigue, physical and psychosocial functioning. **Results:** Self-investigation resulted in significant changes in participants' narratives. Moreover, after self-investigation there was a significant improvement in fatigue, physical and psychosocial functioning for the adolescents with CFS. The patients who completed 12 sessions improved most. At follow-up, the positive effects were maintained. **Conclusions:** Self-investigation enables a move beyond the symptoms of CFS in an individualized, patient centered way. Narrative transformation seems to contribute to improved physical and psychosocial outcome in adolescent CFS. The SCM allows adolescents to discover (for themselves) factors that might cause or perpetuate their fatigue. The results suggest that self-investigation is a useful instrument in the management of adolescent CFS.

INTRODUCTION

Adolescent chronic fatigue syndrome (CFS) is a heterogeneous, medically unexplained condition[1,2]. The main symptom of adolescent CFS is a disabling fatigue that severely affects patients' lives[3]. Although there is growing support for a positive effect of cognitive behavior therapy (CBT)[4,5], a substantial proportion of adolescents with CFS remain severely fatigued and functionally impaired for extended periods of time[6,7]. Therefore, further research into the management of (adolescent) CFS and the customization of its treatment has been urged[8,9].

Recently, there has been an increasing interest in self-management interventions, in which patients are actively involved in their own care[10,11]. These approaches aim at shared expertise and responsibility of patient and professional[12]. Such strategies could prove especially relevant for medically unexplained symptoms, as it is generally agreed that in these conditions treatments actively involving the patients are most effective[13,14]. It has therefore been suggested that the therapeutic focus should go beyond the unexplained symptoms and also take the life-stories of the patients into account[15-17].

Through personal life-stories people try to create coherence in their own history and give their lives meaning and direction[18]. From adolescence onward these narratives are considered to be the building-blocks of identity formation, and help individuals to understand and integrate a variety of positive and negative life-experiences[19]. The forming and transforming of these stories have been shown to bring about improvements in both mental and physical health[20]. Paying systematic attention to the narrative context of adolescent CFS might therefore not only lead to a better understanding of patients' experiences, and provide a more acceptable and effective framework for the management of adolescents' problems based on their own perspectives, but may also entail several health benefits.

The Self-confrontation Method (SCM) is an instrument to assess and change individuals' life-stories, through narrative self-investigation[21]. Personal narratives –far from being neutral descriptions of a succession of events and encounters- are engaged stories of emotionally laden experiences. Therefore, the SCM combines a focus on individuals' verbal reports, with an analysis of the underlying affective and motivational structures. The method's theoretical background[22,23], its clinical use in (adolescent) somatic and psychiatric conditions[24,25], its use to compare groups[26], and its psychometric validity and reliability[27,28], have all been extensively reported. In this small-scale intervention study, the use of the SCM as a self-management tool in adolescent CFS was investigated. It was hypothesized that adolescents' self-investigation of their personal stories would lead to narrative changes and health improvement.

METHOD

Participants

Forty-two consecutively referred adolescents with CFS, who had previously participated in a cross-sectional study[29], were included in this study. These adolescents were all assessed for CFS and the diagnosis, in accordance with the CDC-1994 criteria[30], was either made or confirmed by a pediatrician in the University Medical Center Utrecht, The Netherlands. From a local high-school, twenty-five adolescents without any medical, psychiatric or psychological condition volunteered to participate as a control group. The study was approved by the ethical committee of the University Medical Center, and informed consent was obtained from all participating adolescents and their parents.

Self-investigation procedure

Phase 1, first self-investigation: In order to elicit their life story, adolescents reflect upon three open questions, referring to their past, present and future. In dialogue with a counselor, adolescents formulate their life-stories' most important experiences into written sentences. A self-investigation with an adolescent generally produces 20 to 35 sentences. After the formulation of these sentences, a standard list of 24 affect terms is presented. These affects are subdivided into four scales that are reliable, validated and internally consistent[27]; [1] Positive (P): *joy, happiness, enjoyment, trust, security, and inner calm* (Cronbach's α = .93), [2] Negative (N): *powerlessness, anxiety, shame, self-alienation, guilt, loneliness, inferiority, and anger* (Cronbach's α = .90), [3] Self (S): *self-confidence, strength, self-esteem, pride, energy, and freedom* (Cronbach's α = .91), and [4] Other (O): *caring, love, tenderness, and intimacy* (Cronbach's α = .86). At home on their own, the adolescents indicate on a 6 point Likert-scale (0 = not at all, and 5 = very much) to what extent each affect is related to each particular sentence. Furthermore, in order to get insight into their present overall situation, the adolescents are asked to relate the 24 affect terms to the questions *"How do you generally feel these days?",* and *"How would you like to feel?"*.

Subsequently, the 20-35 sentences that describe meaningful experiences are examined in relation to these affective connotations. Sentences that appear different on a verbal level may show similarity on an emotional level. By computing product-moment correlations between the affective profiles of every sentence, similarities on this implicit level of the personal narrative can be found. In order to find a theme in teenagers' personal narratives, the standardized sentence representing how the adolescents generally feel is used as pivotal sentence. Concentrating on the resemblances between the sentences with a positive correlation ($r > .60$) with the sentence concerning their general feeling, adolescents are asked to summarize the central theme in their personal narratives. In this way, the underlying thematic organization of the life-story is

elicited. In table 1, an abbreviated example of this process is presented for a 16 year old girl with CFS. The first phase is completed in 3 individual sessions of approximately one hour.

Table 1
Abbreviated case example: Establishing a narrative theme

	S†	O†	P†	N†	r‡
General feeling at first self-investigation	1.8	1.0	0.4	4.4	1.00
Ideal feeling at first self-investigation	4.5	4.8	5.0	0.4	-0.83
Self-described sentences					
1) I always feel as if people cannot help me with any of my problems. This makes me anxious and nervous.	1.3	1.0	1.0	4.0	0.95
2) My concentration is really bad. I feel dumb asking questions to adults.	2.0	1.8	0.4	3.9	0.88
3) I often dream that a murderer is chasing me. I hide, but he still finds me.	1.0	0.8	0.8	3.9	0.84
4) I am very jealous and I am always afraid my boyfriend will like other girls better.	2.3	3.5	0.9	4.2	0.72
Self-described resemblance between sentences (1-4)					
(1-2) In my head, I am often quite stressed. I am afraid to look stupid.					
(1-3) I'm in a situation where I'm afraid and alone. I feel helpless and cannot do anything.					
(1-4) Jealousy is a part of me. I'm powerless to change that, just like my CFS.					
Self-described theme					
I feel powerless, frightened and alone. I don't know what to do and I do not think people can help me. It's always very busy in my head and I find it difficult to relax. Therefore I don't sleep well and I can't concentrate. It makes me very tired.					

Note: †S: mean score of self-directed affect; O: mean score of other-directed affect; P: mean score of positive affect; N: mean score of negative affect. Mean scores range from 0-5. ‡r: product-moment correlation with the sentence regarding general feeling.

Phase 2, process-promoting: The central theme emerging from the first self-investigation serves as the guiding principle in this phase. Guided by the counselor, the adolescents start assessing in which daily situations their theme is present or absent, and how they cope with these situations. Subsequently, a gradual move from assessment to change is made. The adolescents begin to create and engage novel situations in order to develop and re-organize their personal narratives. This phase consists of 6 individual sessions of approximately one hour.

Phase 3, second self-investigation: The constancies and changes in the adolescents' personal narratives are evaluated in the form of a second self-investigation. This phase is also completed in 3 individual sessions of approximately one hour.

SCM typology

From the sentences, in combination with their affective organization, six common, psychometrically validated types of experiences, based on the four different affect dimensions (Self, Other, Positive and Negative) can be discerned: *'Strength and Union', 'Aggression and Anger', 'Autonomy and Success', 'Unfulfilled Longing', 'Unity and Love',* and *'Powerlessness and Isolation'*[28]. With these six types at hand it is possible not only to study individual matrices of sentences and affects, but in addition a comparison of group results on the level of emotional experience can be made.

Design

In order to explore two different modes of self-investigation with the SCM in adolescent CFS, the patients were randomly assigned to either 12 or 6 weekly sessions (see figure 1). In the first condition, patients went through all three phases of the SCM under the guidance of a counselor. In the latter condition, the patients only did a first and second self-investigation (phase 1&3) under the guidance of a counselor. For them, between phase 1 and 3 there was a period of approximately six weeks in which they did not have contact with a counselor. In this condition therefore, the adolescents were expected to take more individual responsibility to work with the insights gained from their first self-investigation. All healthy adolescents did a first and second self-investigation (phase 1&3) under the guidance of a counselor. For them as well, between phase 1 and 3 there was a period of approximately six weeks in which they did not have contact with a counselor. Patients agreed not to have other treatment for their CFS during the self-investigation procedure. Questionnaires were used to assess fatigue, physical impairment and psychosocial variables. They were filled in at inclusion, directly after the second self-investigation (at a mean of 4 months), and again at follow-up (at a mean of 10 months after the second self-investigation).

Figure 1
Design and time course

	CFS: 12 sessions	CFS: 6 sessions	Healthy controls
Week 1 *(Phase 1)*	X	X	X
Week 2 *(--)*	X	X	X
Week 3 *(--)*	X	X	X
Week 4 *(Phase 2)*	X	--	--
Week 5 *(--)*	X	--	--
Week 6 *(--)*	X	--	--
Week 7 *(--)*	X	--	--
Week 8 *(--)*	X	--	--
Week 9 *(--)*	X	--	--
Week 10 *(Phase 3)*	X	X	X
Week 11 *(--)*	X	X	X
Week 12 *(--)*	X	X	X

Note. Phase 1: first self-investigation, Phase 2: process-promoting, Phase 3: second self-investigation. X: 1 session, --: no session. Although the aim was to complete the self-investigation procedure in 12 weeks, the mean duration was 4 months.

Outcome measures

Checklist Individual Strength (CIS-20): Fatigue was assessed with the subjective fatigue subscale of the CIS-20[31]. This is a reliable, validated assessment measure, with good internal consistency (Cronbach's α of 0.93) that has previously been used in research of adolescent CFS[4,32].

Child Health Questionnaire (CHQ-CF87): Functional impairment, psychosocial and health related variables were measured with the CHQ-CF87[33]. This is a reliable, validated assessment measure, with good internal consistency and has also previously been used in research of adolescent CFS[34]. The physical role functioning subscale, measuring limitations in school work and daily activities as a result of physical health (Cronbach's α of 0.86), the emotional role functioning subscale, measuring limitations in school work and daily activities as a result of emotional problems (Cronbach's α of 0.90), the behavioral role functioning subscale, measuring limitations in school work and daily activities as a result of behavioral problems (Cronbach's α of 0.71), the bodily pain subscale, measuring severity and frequency of bodily pain (Cronbach's α of 0.85), the general behavior subscale, measuring the exhibition of aggressive, delinquent and immature behavior (Cronbach's α of 0.79), the mental health subscale, measuring a diversity of positive and negative feelings (Cronbach's α of 0.86), the self-esteem subscale, measuring satisfaction with abilities, looks, family/ peer relations and life overall (Cronbach's α of 0.89), and the general health perception subscale, measuring believes concerning health (Cronbach's α of 0.77) were employed[35].

Statistical Analyses

In all statistical analyses SPSS (version 16.0) was used. On the inclusion and outcome variables, group means and standard deviations were calculated. One-way ANOVA's, using post-hoc Bonferroni comparisons, were employed to investigate significant differences between groups. Changes on outcome variables were determined with paired samples T-tests. Potential risk factors for non-improvement were quantified through linear regression with fatigue and physical impairment as dependent variables. The significance level on all analyses was set at $p<.05$ (two-tailed tests).

RESULTS

Thirty-five adolescents with CFS completed the procedure. At baseline their mean age was 16.5 (SD=1.2) years, 85.7% was female, they were all Caucasian, and the mean disease duration was 2.4 years (SD=1.8) years. Sixteen healthy adolescents completed the procedure. At baseline their mean age was 16.3 (SD=0.7) years, all were female, and 93.8% was Caucasian. There were no significant differences in these two groups with regard to age, ethnicity and gender. Seven adolescents with CFS withdrew from the self-investigation procedure. The primary reason for withdrawal was the belief that the self-investigation procedure would not aid in their recovery. At baseline, there were no significant differences between the adolescents with CFS who completed the procedure and those who did not. Nine healthy adolescents withdrew from the self-investigation procedure. The primary reason for withdrawal was that they found the procedure too time-consuming. The healthy adolescents who did complete the procedure were all female and had a significantly higher behavioral and physical functioning score on the CHQ-CF87 ($p<.05$) than those who did not. Five adolescents with CFS did not return follow-up questionnaires. At baseline and at 4 months, there were no significant differences between those participants who returned the follow-up questionnaires and those who did not.

In table 2, the percentages of the different types of experiences in the adolescents' life-stories for those participants that completed the self-investigation procedure are presented. For the most frequently reported types, variables had a normal to near-normal distribution. Both at the first and the second self-investigation there were no significant differences in the proportions of the types of experiences between the CFS/ 6-sessions group, the CFS/ 12-sessions group and the healthy controls. However, in the period between the two self-investigations there were several changes within the three groups. At 4 months, both the CFS/ 6-sessions group and the CFS/ 12-sessions group reported significantly fewer experiences of powerlessness and isolation. In addition, the CFS/ 12-sessions group also reported significantly fewer experiences of unfulfilled longing and unity and love, and described

Table 2
Proportion (%) of types of experiences within adolescents' total narrative systems

	Scores, Mean (SD)		Change, Mean (SD)
	0 months	4 months	0-4 months
Strength and union[1]			
CFS: 6 sessions (n=17)	13.0 (13.6)	10.9 (14.5)	-2.0 (13.3)
CFS: 12 sessions (n=18)	13.2 (12.2)	16.2 (18.5)	3.0 (15.7)
Healthy controls (n=16)	8.4 (11.5)	16.8 (19.7)	8.4 (12.8)*
Autonomy and succes[2]			
CFS: 6 sessions	26.3 (18.7)	30.2 (19.6)	3.9 (14.7)
CFS: 12 sessions	24.4 (10.9)	43.8 (17.5)	19.3 (14.4)*
Healthy controls	32.2 (15.4)	32.7 (18.0)	0.5 (17.0)
Unity and love[3]			
CFS: 6 sessions	2.2 (4.0)	2.9 (4.7)	0.7 (3.7)
CFS: 12 sessions	5.4 (8.2)	1.2 (2.6)	-4.2 (6.6)*
Healthy controls	3.3 (4.9)	1.6 (2.5)	-1.6 (4.2)
Powerlessness and isolation[4]			
CFS: 6 sessions	17.0 (15.2)	10.9 (13.2)	-6.1 (7.7)*
CFS: 12 sessions	13.4 (6.8)	6.8 (7.8)	-6.5 (11.4)*
Healthy controls	12.0 (12.2)	10.5 (11.6)	-1.5 (10.2)
Aggression and anger[5]			
CFS: 6 sessions	3.1 (4.9)	2.3 (4.2)	-0.8 (6.1)
CFS: 12 sessions	4.3 (8.4)	3.3 (6.5)	-1.0 (7.9)
Healthy controls	3.3 (6.0)	1.2 (2.2)	-2.0 (5.0)
Unfulfilled longing[6]			
CFS: 6 sessions	5.7 (9.7)	4.0 (8.9)	-1.7 (3.8)
CFS: 12 sessions	9.1 (10.3)	4.1 (5.0)	-5.0 (7.1)*
Healthy controls	3.5 (4.4)	1.6 (3.3)	-1.8 (4.0)

Note. See [28] for psychometric data regarding types of experiences. [1]High self-directed affect (S), high other-directed affect (O), high positive affect (P), low negative affect (N). [2]High S, low O, high P, low N. [3]Low S, high O, high P, low N. [4]Low S, low O, low P, high N. [5]High S, low O, low P, high N. [6]Low S, high O, low P, high N. *Indicates significant change on the $p<.05$ level.

significantly more experiences of autonomy and success at the second self-investigation. At 4 months the healthy adolescents reported significantly more experiences of strength and union.

In table 3, the scores on the measures of fatigue, physical and psychosocial functioning are presented. Variables had a normal to near-normal distribution. At baseline, the two CFS groups did not differ from each other, but had significantly worse fatigue, physical functioning, bodily pain, mental health, self-esteem and general health perception scores in comparison to the healthy adolescents. At 4 months, both CFS groups showed a significant improvement on the fatigue, physical functioning, mental health and general health perception scales.

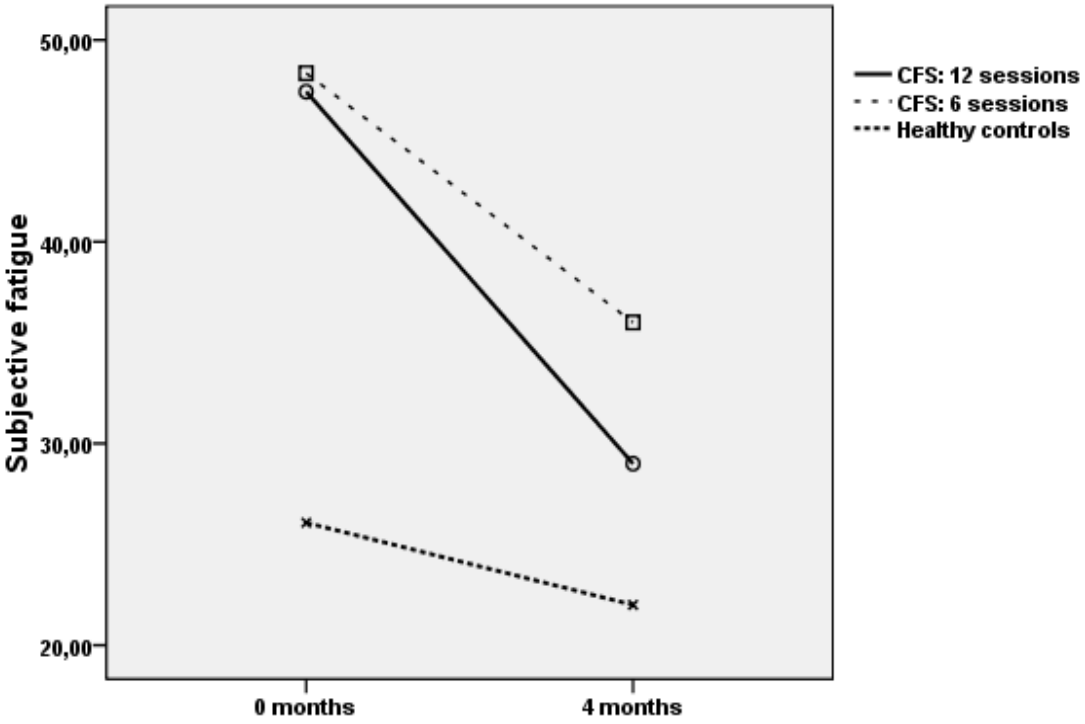
Table 3
Fatigue, physical and psychosocial functioning

	Scores, Mean (SD)			Change, Mean (SD)
	0 months†	4 months†	14 months‡	0-4 months
Subjective fatigue[1]				
CFS: 6 sessions	48.4 (7.5)[a]	36.0 (12.2)[a]	34.7 (12.6)[a]	-12.4 (9.4)*
CFS: 12 sessions	47.4 (7.6)[a]	29.0 (12.4)	29.2 (12.0)	-18.4 (12.6)*
Healthy controls	26.1 (10.9)[b]	20.7 (10.9)[b]	21.3 (11.2)[b]	-5.4 (7.1)*
Role functioning: Physical[2]				
CFS: 6 sessions	34.6 (24.8)[a]	59.5 (29.1)[a]	69.2 (33.4)[a]	24.8 (32.5)*
CFS: 12 sessions	36.3 (24.3)[a]	79.3 (25.5)	81.7 (18.4)	43.0 (29.7)*
Healthy controls	91.1 (17.4)[b]	96.3 (10.0)[b]	97.0 (8.9)[b]	5.2 (19.2)
Role functioning: Emotional[2]				
CFS: 6 sessions	68.0 (33.5)	81.0 (25.7)	75.2 (25.3)	13.1 (28.9)
CFS: 12 sessions	60.0 (40.5)	85.9 (19.5)	88.2 (14.4)	25.9 (38.4)*
Healthy controls	86.7 (17.4)	92.6 (10.0)	87.4 (25.8)	5.9 (15.1)
Role functioning: Behavioral[2]				
CFS: 6 sessions	85.0 (24.2)	94.8 (13.1)	92.3 (12.3)	9.8 (25.4)
CFS: 12 sessions	78.5 (34.7)	94.1 (10.2)	93.5 (13.1)	15.6 (33.3)
Healthy controls	97.0 (6.6)	98.5 (3.9)	92.6 (25.8)	1.5 (7.1)
Bodily pain[2]				
CFS: 6 sessions	41.8 (26.0)[a]	53.5 (21.2)[a]	52.3 (32.2)[a]	11.8 (28.1)
CFS: 12 sessions	35.3 (22.3)[a]	58.0 (22.4)	55.9 (28.7)[a]	22.7 (22.5)*
Healthy controls	72.7 (21.2)[b]	76.7 (20.6)[b]	83.3 (15.0)[b]	4.0 (13.5)
Mental health[2]				
CFS: 6 sessions	58.8 (16.1)[a]	65.2 (10.0)	66.3 (20.8)	6.3 (11.8)*
CFS: 12 sessions	54.5 (14.5)[a]	67.3 (11.3)	67.4 (13.5)	12.8 (15.0)*
Healthy controls	71.7 (10.4)[b]	74.4 (12.5)	72.0 (16.7)	2.7 (6.6)
Self esteem[2]				
CFS: 6 sessions	55.4 (12.0)[a]	61.7 (13.2)[a]	64.8 (19.1)	6.3 (15.0)
CFS: 12 sessions	58.3 (12.6)[a]	72.3 (12.5)	67.8 (12.8)	14.1 (12.0)*
Healthy controls	71.1 (8.0)[b]	76.3 (11.7)[b]	71.4 (15.1)	5.2 (8.7)*
General health perception[2]				
CFS: 6 sessions	42.9 (15.8)[a]	51.3 (12.8)[a]	53.0 (19.2)[a]	8.4 (12.0) *
CFS: 12 sessions	36.5 (18.8)[a]	50.7 (22.0)[a]	49.2 (22.1)[a]	14.2 (20.2)*
Healthy controls	77.7 (10.9)[b]	79.7 (13.5)[b]	78.2 (12.6)[b]	2.0 (9.8)

Note. †CFS: 6 sessions (N=17), CFS: 12 sessions (N=18), Healthy controls (N=16) ‡CFS: 6 sessions (N=13), CFS: 12 sessions (N=17), Healthy controls (N=16). There were no significant changes during the follow-up period, so these change-scores are not indicated in the table. [1]Subjective fatigue subscale (CIS-20): score ranges from 8-56; a high score indicates a high level of fatigue. [2]CHQ-87 scores range from 0-100; higher scores indicate better functioning. [a,b]In the columns, different superscripts indicate significant group differences on the $p<.05$ level. *In the rows, indicates significant change on the $p<.05$ level

For a general illustration of these effects; see figure 2.

Figure 2
Subjective fatigue



In addition, the CFS/ 12-sessions group also showed a significant improvement on the emotional role functioning, bodily pain and self-esteem scales. Furthermore, except on general health perception, after the second self-investigation the CFS/ 12-sessions group no longer differed from the healthy controls on any of the scales. In contrast, while the CFS/ 6-sessions group showed several significant improvements, at 4 months they continued to differ from the healthy controls on the fatigue, physical functioning, bodily pain, self-esteem and general health perception scales. After the second self-investigation the healthy adolescents showed a small, but significant improvement on the fatigue and self-esteem scores. At follow-up 10 months after the second self-investigation, there had been no significant changes on any of the scores and the positive effects were maintained.

Linear regression analysis indicated that only a high fatigue score at baseline was a risk factor for a high fatigue score (t=3.22; p=.01), and a low physical functioning score (t=2.16; p=.05) at 4 months.

DISCUSSION

The results seem to indicate that, in adolescent CFS, significant improvement in health and psychosocial variables is accompanied by a mechanism of narrative transformation. In SCM-theory it is generally assumed that positive change is initiated by a reduction of experiences of powerlessness and isolation[36]. This process can be witnessed in both CFS groups. Further improvement is usually in the direction of a predominance of experiences of autonomy and success. In the course of counseling, this development towards more autonomy and activity often gets (temporary) priority over relation-oriented experiences, such as unity and love. This specific kind of process seems to underlie the major improvements in the CFS/ 12-sessions group.

Somewhat to our surprise we did not find (initial) differences in the types of experiences in the life-stories of adolescents with CFS and healthy controls. Furthermore, because of the infrequent reporting of some narrative types (e.g. unity and love & aggression and anger), a normal distribution of these variables was not guaranteed. We believe this to be due to the modest size of the sample described. Moreover, even though Bonferroni corrections for the ANOVA analyses were made, a rather large number of statistical tests was done, increasing the risk of type 1 errors. Finally, as this was a small-scale study, we did not include a waiting list control group. Though patients had been symptomatic for a long time, it cannot be completely ruled out that improvements were partly spontaneous and/ or the result of the attention of a health care professional. The results therefore require reproduction in a larger-scale randomized controlled trial.

This is the first study reporting on the use of narrative self-investigation in adolescent CFS[37,38]. The female to male ratio was in accordance with research findings, the mean age of participants did not have a wide range, and only adolescents diagnosed with CFS according to the CDC-1994 criteria were eligible for the study. Like in previous studies, few risk factors for continued fatigue and physical impairment were identified[39]. The percentage of participants who withdrew from the study is similar to other studies into the management of adolescent CFS with CBT[4]. Both CBT and narrative self-investigation show positive effects in the self-reported health status of adolescents with CFS. The difference between both methods is primarily one of focus. While CBT addresses the maintenance of symptoms through a change of cognitive processes[40], the SCM intends a move beyond the symptoms by eliciting emotions and establishing a personal narrative theme.

Personal narratives are unique. By incorporating the broader context of patients' affective life-stories into the management of adolescent CFS the therapeutic focus is sharpened. Furthermore, self-investigation offers adolescent patients the opportunity to explore those elements of their problems –which might not exclusively be fatigue or physical impairment- that they themselves find most important and feel they can influence. A continuing dialogue with a counselor during this process seems to stimulate a comfortable transition from assessment to change.

This investigation of their own experience and the changes in their life-stories is reflected in significant improvements in fatigue, physical and psychosocial functioning in both CFS groups. At follow-up, this positive effect was maintained. At 4 months as well as at 14 months, there was a significant difference in improvement between the two CFS groups. Those patients who had 12 sessions in the self-investigation procedure showed considerably more narrative changes and improvement on outcome measures than those who had only 6 sessions. A more intensive counseling procedure is therefore advisable.

CONCLUSION

The SCM is fundamentally a collaborative process between the adolescents and their counselor, through which the patients are challenged and empowered to be experts of their own life and experiences. The dialogue supporting the process of self-investigation enables the counselor to move beyond the symptoms of CFS, and to address previously unexpressed problems in an individualized, patient centered approach. It allows the adolescents to discover for themselves factors that might cause or perpetuate their fatigue. The results are positive and suggest that enabling adolescent patients with CFS to participate in their own treatment stimulates narrative transformation and health improvement.

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Chapter 6

General discussion: Understanding self-experience in adolescent chronic fatigue syndrome

People seek help from doctors because they suffer from complaints such as pain, discomfort or fatigue. Usually, physicians will understand these symptoms as the patients' subjective experience of an underlying disease. They will then try to make a diagnosis of this illness, and prescribe a treatment based on this assessment[1]. Often however, a clear pathology is not found. When physical symptoms cannot be defined in conventional terms of disease, such complaints will commonly be referred to as part of a medically unexplained illness. In recent years these conditions, such as irritable bowel syndrome, tension headache, fibromyalgia and chronic fatigue syndrome (CFS), have become a growing concern for health care. Although unexplained, they are now quite common, and often chronic in nature[2]. In addition to this, these syndromes are usually associated with significant distress for patients, and the use of clinical resources and the economic costs are consistently increasing[3].

Some of the major challenges these illnesses pose are of a conceptual nature. The functional somatic syndromes confront medical science with the limitations of the traditional paradigms, through which such remarkable progress in the understanding and treatment of many diseases has been made. In this final chapter, CFS will be presented as a typical example of the medically unexplained illnesses. The major conceptual problems in this syndrome will be discussed. Subsequently, it will be argued that recent theoretical developments, concerning the dialogical and bodily nature of the self and self-experience, might help to overcome some of these problems. After considering two case-histories of adolescents with CFS, it will then be evaluated whether such insights might lead to a better understanding of some main features of the syndrome. Finally, some clinical and research implications of this perspective on CFS are discussed.

Chronic fatigue syndrome

CFS is a syndrome of unknown origin. It is characterized by prolonged, severely disabling fatigue and it is associated with symptoms such as myalgia, headache, sleep disturbance, swollen lymph nodes, pain, and memory- and attention difficulties. The fatigue and related symptoms are not the result of ongoing exertion, are not substantially alleviated by rest, and seriously limit physical and social functioning[4].

Etiological studies of CFS have, at first, mainly focused on somatic pathology. Viral or bacterial infections, immune system dysfunctions and disturbances of the neuroendocrine or neurotransmitter systems have all frequently been investigated as plausible causes of CFS[5,6]. However, although sometimes minor changes in a proportion of patients have been found, it has generally been concluded that no consistent evidence of somatic abnormalities can be demonstrated. Furthermore, it is unclear whether biological changes, if

any, are primary or secondary to the onset of CFS. As it is often the case in biomedical practice when patients present with bodily symptoms and no physical pathology is found, it is then assumed that the symptoms must be psychopathological in origin[7].

Consequently, the psychiatric and psychological status of patients with CFS has also received much attention[8]. Numerous studies have investigated the role of depression, anxiety, alexithymia and personality disorder, while other studies suggested that in CFS we are dealing with a specific form of somatization, hypochondria or conversion[9]. Some studies concluded that psychopathology was prevalent among patients with CFS and that these psychiatric conditions, in some cases, predated the development of the syndrome. Other studies however, concluded that psychiatric disorder, if any, mostly manifested itself after the onset of the fatigue and therefore was more likely to be a consequence, rather than a cause of the syndrome.

So, on the one hand, there are those who believe that the symptoms of CFS are caused by still unknown biological mechanisms. The lack of findings of a clear organic cause on the other hand, leads others to refer to CFS as a psychopathological condition. In that case, CFS is generally thought of as being 'all in the mind'. However, no consistent evidence for either of these claims has been found[10]. Consequently, as bodily symptoms from a biomedical point of view are considered as either a manifestation of physical pathology, or as a manifestation of mental illness, medical science is faced with a conceptual problem. In the continued absence of a definite pathology underlying the patients' symptoms, the question thus needs to be explicitly raised; what kind of phenomenon are we actually trying to explain? Are fatigue and functional impairment truly the central problems of CFS?

Beyond the symptoms

In recent qualitative studies, patients with CFS indicate that while they are severely fatigued and impaired, this is not necessarily their only, or even their main concern. They describe a pervasive feeling that their bodies no longer hold the capacity for involvement with other people, leading to social marginalization. This reduced interaction in combination with a decrease in physical activity is felt to lead to a confounding of affect and a lack of opportunities to express emotion. Furthermore, they point to the fact that they feel that biographical disruptions have led to a fundamental loss of self, and they report that negative personal experiences lead to self-doubt and a further withdrawal from contact with others. They also report the overwhelming sense of no longer being in control of their own lives, and feeling as being reduced to an inferior version of their former self. They feel as if their selves are continually diminishing, spiraling downward, and as if they are dislocated from the rhythms of their lives[11-17].

From these descriptions, it becomes apparent that underlying the symptoms and clinical features of the syndrome there is almost without exception the emotional experience of a disrupted and isolated sense of self. At the core of the suffering in CFS there seems to be a profound alteration of self-experience and identity. A major focus of attention should thus become how this phenomenon of self-alteration can be conceptualized.

In the currently predominant understanding of CFS, a biopsychosocial approach is often combined with a cognitive-behavioral model. In these models, it is assumed that multiple cognitive and behavioral factors interact with physical and social aspects to result in symptoms[18,19]. For example, it is proposed that cognitive characteristics account for physical and psychosocial disability in CFS, that patients are characterized by problems in thought specific to their somatic experience, that rigidly held beliefs act to defend patients against low self-esteem, and that cognitive idiosyncrasies cause the perception and experience of fatigue in the syndrome[20-23].

In recent years, several conceptual concerns with regard to these models have been expressed. Cognitive-behavioral models (at least on a theoretical level) rely heavily on the idea that our emotions and behavior are mediated by our thoughts and beliefs[24,25]. However cognitive-behavioral models tend to be overly concerned with symptoms on an explicit cognitive level, and tend to neglect underlying, implicit affective meanings[26,27]. Furthermore, while the biopsychosocial and cognitive-behavioral models have been proclaimed as a paradigm shift, because they incorporate biological, as well as psychological and social elements in a multidimensional understanding of CFS, they continue to incorporate some of the conceptual presuppositions of the biomedical model[28]. The different levels of these models are commonly still regarded as causally interacting factors and there is little attention to their intrinsic inseparability[29]. Therefore, the dichotomies between mind and body, and between the inner person and the outside, physical and social world appear to remain intact[30].

Certainly, the biopsychosocial and cognitive-behavioral models acknowledge the centrality of the patient as the focal point in which biological, psychological and social factors come together[31]. However, they have primarily been developed to enable the *objective* study of the pathological causes and consequences of illness on the one hand, and the treatment of the patients' dysfunctional symptoms on the other. As theoretical models, they are therefore less suitable to conceptualize *subjective* self-experience in the condition. In what follows, it will be argued that an incorporation of current dialogical and embodied conceptualizations of the self allow a more integrative view on persons with CFS, less focused on pathology and symptomatology, and more sensitive to the inseparable relational, narrative, emotional and bodily dimensions of self-experience.

The dialogical self

In recent decades, there has been an increasing recognition of the fact that humans do not primarily encounter and engage the world on the basis of their beliefs and thoughts about themselves, their surroundings and others. Instead, it is argued that persons evaluate and structure their experiences through contextualized narratives. They assess their behavior and interactions, interpret themselves and others, and articulate what they believe to be important in a narrative form[34]. As their experience of the world is continually changing, these narratives further structure temporality and bring unity to their past, present and future[35]. These narratives however, do not have a fixed composition, but can only be accomplished in a continuous dialogue with others. Our sense of self is -to a large extent- considered to be based on these stories. These self-narratives are constructed as we live in the social world and are therefore always interwoven and entangled with other people's stories.

This narrative view of the self and self-experience has been further developed by dialogical self theory[36,37]. While acknowledging the human striving toward a single and coherent life story, dialogical self theory at the same time points to the fact that narrative identity emerges from a plurality of perspectives of the self, or 'self-positions'. According to this theory, the self is constructed in open dialogue not only with others, but also between different complementary and opposing perspectives within the self. The dialogical self is understood as a dynamic multiplicity of fluctuating perspectives of the self. It is assumed that such self-positions, of which some are more dominant than others, entail a multiplicity of narratives. Some of these narratives will be long and complex, others shorter and more straightforward, some will be more in line with culturally predominant themes, others more idiosyncratic. Our self and our stories are able to evolve and adapt to the challenges of life through a continuous renewal and shifting in the dialogues between, and the organization of, our many self-positions. Our sense of unity and coherence of 'the' self does not come from adopting a single, unitary perspective, but instead results from a continuous re-ordering of interacting aspects of our selves through dialogues with ourselves and others[38]. It is assumed that a healthy organization of self-positions implies flexibility in an individual's self-organization, and the potential to move from one position to another. In order to respond adequately to changing contextual demands, a person therefore needs to be able to employ an emotionally varied repertoire of self-positions[39].

Now, such a dialogical view of the self, to a large extent, seems more sensitive to the intrinsic and inseparable relation between individuals, and their social and cultural context. However, the question can be raised how people's experience of themselves as (relatively) coherent, stable and continuous is possible, when identity is regarded as emerging from a multiplicity of positions and a polyphony of voices[40]. Additionally, this notion of the self runs the risk

of becoming overly dependent on our reflective story-telling abilities. In that case, it could be suggested that it fails to conceptualize what it is like to experience the world affectively and pre-reflectively[41]. Furthermore, it could also be argued that such a notion of the self still seems to have little attention for the bodily dimensions of subjective and intersubjective experience. Therefore, its relation to current bodily notions of the self will briefly be considered in the next section.

The bodily self

Presently, there is a renewed interest and appreciation of phenomenological views on the embodied dimensions of self-experience[42,43]. The notion is now becoming more commonplace that the self is fundamentally constituted and developed by our practical agency, and engagement with the world, against an always present pre-reflective background of inarticulate bodily schematizations. These body schematic processes are nearly automatic, and involve the sensorimotor capacities and proprioceptive- and interoceptive awareness that underlie intentional action[44].

Narrative identity is intrinsically related to this bodily experience, and is characterized by a continuous back and forth movement, between reflective and pre-reflective experience. Our body, its movements and perceptions, play a key role in our narrative sense of self because, in a way, the body provides our 'path' through space and time and as such underpins and secures our familiarity with the world[45]. Our body is always actively involved with its surroundings and the qualitative sense of our embodiment pervades, integrates and unites the polyphony of the self's diverse narratives. Furthermore, most of our general self-experience has an obvious bodily dimension[46]. Our perceptual experience, for instance, appears to have an irreducible grounding in the body as it provides the tacit reference point on which we base our judgments of distance, size, movement and speed of objects in the physical world[47]. In intersubjective perception, our body from birth acquires the capacity to 'transpose' the perceived actions, movements, gestures and intentions of others into its own motor schemes and we sense the emotion of others in the movement and expression of the other's body[48]. In intersubjective interaction we are guided by these movements and expressions of the other and mirror them, and vice versa[49].

Furthermore, the implicit and integrative functioning of the body in everyday interaction, understanding and perception embeds the self in an immediate and irreducible relation to the world[50]. Our sense of being seems to stem from, and to be constituted by, a background of bodily felt states such as excitement, hunger, anxiety, sexual craving, anger, joy and impatience. As such, these embodied states of the self appear to constitute an anchor that ties us to the world and opens it up -or closes it off- as a meaningful realm of dialogue

and interaction. Our moods and emotions seem to be a prerequisite for a successful 'relation' of the self to the world. They tune us to the world by constraining and structuring our possibilities, making some options for action appear as more relevant, meaningful and attractive, than others[51,52]. Probably only those events and situations that are thus designated as significant make it into our self-narratives[53]. The personal narratives constituting self-identity would thus never be neutral descriptions of a succession of events and encounters, but would instead always be engaged stories of emotionally laden experiences.

Although discussed only very concisely, a framework based on a dialogical and embodied notion of the self might be able to provide an integrated account of individuals' relational, narrative, emotional and bodily self-experience. On this account, one could say that humans are primordially bodily and socialized beings who understand, communicate and interact with themselves, and others, through narrative self-conception; the dialogical self is necessarily a bodily self. But how do these reflections on the bodily and dialogical nature of the self relate to CFS?

The bodily, dialogical self in adolescent CFS

Let us consider two cases of chronically fatigued and functionally impaired adolescent patients, in order to clarify possible ways in which a bodily, dialogical concept of the self might shed more light on the experience of CFS:

Case I: John is a 16-year old adolescent boy, who became severely ill after an Epstein-Barr virus infection. The main symptom of such an infection is severe fatigue and it can last for a significant time, in this case almost 2 months. As there is no uniform medical cure for this infection, the remedy in his case consisted primarily of sleep and rest. During this period John did not attend school, limited physical and social activity to a minimum, and remained mostly indoors. After this period however, although there were no longer any viral markers for a persisting infection, John's fatigue intensified and his symptoms progressed to the point where they also included headaches, stomachache, and severe pain in his limbs. When he visited our pediatric hospital it had been six years since the initial infection and in this period he had been diagnosed with CFS, and had barely felt able to attend school. He was then asked to participate in this thesis' study into narrative counseling for chronically ill adolescents. At the start of this, he indicated that he felt thoroughly bored being at home all the time. He also expressed the feeling that he had become too dependent on his parents, and that he had very few experiences of autonomy or success. He started a lot of projects, but was never really able to finish any of them. When he felt sad or angry, he withdrew to his room in order not to bother others. He spent most of his time playing online, multi-player computer games. In his own

words; *"I play World of Warcraft because then I can retreat to another world and I don't have to think about the real world."*

Case II: Alice is a 15-year old adolescent girl, who gradually developed symptoms of fatigue, muscle- and joint pain, and concentration problems during a period in which she changed to a new school. At the same time, her parents were going through a complicated divorce. When she visited our pediatric hospital it had been 2 ½ years since her symptoms began, and in this period she had been diagnosed with CFS. During this time she still attended school, and in fact focused most of her remaining energy on her school performance. She was also referred to the counseling program, and soon indicated that she always wanted to help other people with their problems and had felt very powerless and isolated when she came to realize that there was nothing she could do to save her parents' marriage. At present, she felt that her mother had become completely self-absorbed, and that there was very little room for her needs and wishes. She also felt that her siblings got all of her mother's attention. She described that, instead of building up unrealistic expectations of her mother, she had taken up a stance of resignation, and had withdrawn from that part of her social and emotional life in order to protect herself from further disillusions. In addition, she recounted that -around the same time as her parents were divorcing- she witnessed some close relatives involved in a serious car accident. She had always been highly sensitive of her body and since that moment she felt like she couldn't use her legs when something unexpected happened, and she quickly became fatigued when she experienced adversity.

At the time they were seen in our hospital, in neither of these cases was there any clear evidence of enduring pathology, either 'physical' or 'mental'. In that sense their CFS was unexplained. However, both cases entailed a significant change of the adolescents' normal experiences of their selves, and their usual ways of being in the world. To both adolescents it felt as if their bodies had turned into a material object[54]. Instead of opening up the world and its full scope of possibilities for creative activity, social interaction and dialogue, their bodies as well as their narrative sense of identity had come to a standstill and seemed to close the adolescents off from their world and with it, to a large extent, their sense of potentiality[55,56]. The overwhelming experience of fatigue and related symptoms as such seemed to reflect a loss of an immediate, spontaneous and intimate basic sense of self, and a corresponding alienation from the tacit, commonsensical meanings of the physical and social world.

In the first case, a primarily 'bottom-up' understanding of this phenomenon seems plausible. The infection and the subsequent prolonged lack of physical and social activity had quite literally led to a severe disturbance

in the adolescent's tacit, bodily dimension. Without the diverse emotional responses and interruptions solicited by a whole range of positive and negative experiences there was very little to break or disturb a narrow narrative coherence, and an ongoing identical interpretation of events[51]. In the sustained absence of other stories to tell and self-positions to occupy, one position (or group of positions) *I-as-sick*, *I-as-alone*, *I-as-fatigued*, or *I-as-completely dependent on others* became dominant, rigid, internally consistent and almost impervious to change. In the second case, a primarily 'top-down' explanation of the phenomenon of self-alteration seems more likely. In this case, the repertoire of positions the adolescent could adopt (e.g. *I-as-loving-my mother, I-as-a-daughter, I-as-helping others*) in meaningful communicative interactions with important people in her life (e.g. her mother, her former classmates), had quite abruptly diminished. This reduction in the accessibility of some positions entailed a lack of a whole range of possibilities for emotional experience and expression, and thereby a related narrowing of narrative identity[57]. This loss of dialogical capacity and the subsequent restrictive coherence of her self translated into a rigid and objectified body-experience. Her narrative coherence stopped to be a continuous process of ongoing dialogue and interaction, instead resulting in an inflexible physical self-positioning and a disruption of bodily potentiality.

It seems evident that all persons with CFS are affected in their own particular way and that the symptoms are realizable in multiple manners[58]. This also explains the lack of any findings of a consistent pattern of 'abnormalities'. However, underlying all the different manifestations of the symptom of fatigue, there seems to be a common phenomenon of self-alienation in CFS. Over the course of time, the bodily, dialogical self of individuals with CFS looses its familiarity and attunement with itself, and the world. As a consequence, it turns into an overly coherent 'monological' self with foremost a hyperreflective, representational relation to its own objectified body and others. This loss of dialogical capacity and tacit bodily familiarity disturbs patients' sense of self, and intensifies symptoms. At the same time, a worsening of the symptoms might well further the process of dialogical collapse and hinder bodily attunement.

As CFS involves a profound alteration of bodily and dialogical self-experience, therapeutic focus should go beyond the symptoms and address the underlying sense of self. Recovery from CFS will most likely involve helping patients with narrative transformation and the establishment of a renewed bodily sense of their identity[56, 59]. One way to engage patients' selves in treatment would be to start from the narratives patients tell about themselves[60]. The self is not only a given narration that can be studied by patients themselves, but also something that can be transformed in dialogue with a counselor or therapist. However, not only patients' self-narratives should be a focus of therapeutic attention, but also the emotions that the patients relate to

these stories, and the positions within the bodily, dialogical self from which they are told should be investigated[61]. Narrating their lives from different self-positions will allow patients with CFS a recasting of their past, and a re-visioning of their future. In this way, through external dialogue, communications between self-positions can be recovered and new feelings, possibilities and perceptions can be opened[62]. In the counseling of persons with CFS, patients should be helped, as self-investigators, in retelling and renewing their affective life-stories, thereby opening up the possibility of once again engaging fully and directly with the social and physical world.

Remaining questions

This discussion was an attempt to outline a new conceptual framework for the understanding of self-experience in adolescent CFS. Of course this attempt, in the brief space of this chapter, can only be provisional. Several research and clinical questions with regard to self-experience in adolescent CFS remain. The most important would seem to be:

1) Severe fatigue is a common phenomenon during adolescence[63]. However, not every fatigued adolescent develops CFS. Furthermore, many adolescents suffer from infections (as John), or experience psychosocial difficulties (as Alice) and do not develop CFS either. As CFS seems to be characterized by a serious self-alteration, the question remains which individuals are vulnerable and under what particular circumstances this change comes about.
2) Alterations in the bodily, intersubjective and dialogical dimensions of self-experience have recently been described in other somatic, psychiatric and medically unexplained conditions as well[48,64-66]. A related issue is therefore, in what way the transformation of self-experience in CFS differs from those described in other illnesses.
3) Narrative self-investigation primarily addresses issues on the relational, narrative and emotional levels of self-experience. To optimalize self-management in adolescent CFS a new challenge will be to find ways to integrate the bodily level of self-experience into treatment as well, as this would seems to further contribute to a favorable outcome[67,68].

Conclusion

The main concern of this thesis was to understand self-experience in CFS. Rather than providing a novel explanation of the etiology of the syndrome it attempted to clarify a common, yet heterogeneous experiential phenomenon underlying the symptoms of the condition. This discussion started from the assumption that the current explanatory models of CFS are based on a too narrow, and overly reflective account of what it means to be a person. By seeing the symptoms of individuals with CFS primarily as stemming from dysfunctional cognitions or other pathologies, the emphasis is taken away from patients' pre-reflective, practical, bodily and intersubjective being in the world. It was argued that the overwhelming experience of fatigue and impairment corresponds to a profound alteration of self-experience, through a collapse of dialogue, a disruption of affective potentiality, and a loss of connectedness with one's own physicality as a lived-body. It was argued that treatment inspired by such a dialogical and bodily concept of the self in CFS should focus primarily on narrative transformation, and a regaining of the patients' sense of attunement with their physical and social being in the world.

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Summary

SUMMARY

A striking feature of the chronic fatigue syndrome (CFS) is the lack of consistent findings of pathology. While there has been a continuing search for the etiology and pathogenesis of the syndrome, there has been relatively little interest in a clarification of the experiential phenomena underlying its symptoms. The first aim of this thesis was to come to a better understanding of adolescent patients' self-experience. The second aim was to test whether counseling based on their self-experience would lead to an improvement of their health.

Chapter 1 describes the background against which the thesis' main study was initiated. CFS has always been the topic of debate between patients, caregivers and scientific researchers. One source of controversy was the assumption that their is something 'special' about the patients with CFS; they seem to be a certain type of person, and their self-experience seems to differ from healthy individuals and patients with distinct somatic illnesses. To investigate such assumptions a study of the personalities of patients seems the most logical approach. The main aim of this chapter was therefore to review the available personality research in CFS. In these studies, there is some evidence that patients with CFS score higher on particular personality traits than healthy controls do. However, when compared to patients with other chronic illnesses, the findings of specific personality differences is less obvious. Overall, on the trait level of personality there seem to be very few consistent findings. Therefore it was hypothesized that new insights into personality and self-experience in CFS might be gained by systematically studying the self-positioning and affective self-narratives of patients with the syndrome. With regard to the study's design, a longitudinal research approach was suggested. CDC-criteria for CFS should be strictly applied. Furthermore, the study should include age and sex matched healthy individuals, and patients with a somatic illness as control groups.

Chapter 2 describes some of the baseline measurements of this thesis' research, and also introduces the study's main theoretical assumptions with regard to the self and self-experience. In dialogical self theory, narrative identity is seen to emerge from a plurality of self-positions. According to this theory, the self is constructed in dialogue not only with others, but also through exchanges between different complementary and opposing

perspectives of the self. In this chapter, the findings with regard to the self-positioning of 42 adolescents with CFS, 37 adolescents with Juvenile Idiopathic Arthritis (JIA) and 23 healthy teenagers are presented. It was found that adolescents with CFS report severe physical difficulties and psychosocial problems. They positioned themselves as significantly less strong and more unwell in their social relationships in comparison to healthy adolescents, and patients with JIA. There was a strong indication that positions relating to illness were overly dominant.

Chapter 3 describes a further investigation of these findings, and addresses the patients' affective life-stories underlying the self-positioning as reported in the previous chapter. Furthermore, the study's primary research instrument, the self-confrontation method (SCM) is introduced. The SCM is an instrument to assess and change individuals' self-experience, through narrative self-investigation. It combines a focus on individuals' verbal reports, with an analysis of the underlying affective and motivational structures. In this chapter, it was found that adolescents with CFS report significantly less positive experiences of autonomy and success, in comparison to healthy adolescents. In addition, adolescents with CFS described significantly more negative experiences of powerlessness, isolation, and unfulfilled longing compared to healthy teenagers and adolescents with JIA. It was therefore regarded of primary importance to address these issues in clinical settings

Chapter 4 presents the results of a separate follow-up study into the prognosis of adolescent CFS after regular treatment. In the main research, described in this thesis, no waiting list control group was included. Therefore in this chapter, the symptomatic and educational long-term outcome, healthcare use, and risk factors for non-recovery in adolescent CFS are described. A previous cohort of 60 adolescents seen in the University Medical Center Utrecht, The Netherlands, was followed up at a mean of 2.2 years (SD, 1.6). About half of the adolescents had recovered from CFS at follow-up. However, the other half was still severely fatigued and physically impaired, indicating that despite intensive health care use a substantial proportion of adolescent patients with CFS remain symptomatic for a considerable time. A poor outcome was associated with substantial bodily pain, and low mental health, self esteem and general health. It was therefore concluded that there

was a persisting need for the further customization of existing treatment, and for the study of additional interventions.

Chapter 5 presents the results of the main study into narrative self-investigation in adolescent CFS, and the longitudinal changes in patients' self-experience. The adolescent patients with the syndrome described in chapters 2 and 3 were randomly assigned to either 6 or 12 sessions with the SCM. The healthy youngsters were assigned to 6 sessions. Self-investigation resulted in significant changes in the participants' affective self-narratives. Moreover, after self-investigation there was a significant improvement in fatigue, physical and psychosocial functioning for the adolescents with CFS. The patients who completed 12 sessions improved most. At follow-up, the positive effects were maintained. It was therefore concluded that self-investigation not only enables change in the symptoms of CFS, but also of the underlying self-experience of adolescents, in an individualized, patient centered manner. Narrative transformation seems to contribute to improved physical and psychosocial outcome in adolescent CFS. The SCM allows adolescents to discover (for themselves) factors that might cause or perpetuate their fatigue. The results suggest that self-investigation is a useful instrument in the management of adolescent CFS.

Chapter 6 is the general discussion. In this chapter an attempt is made to outline a new conceptual framework for the understanding of self-experience in adolescent CFS. It started from the assumption that the prevalent explanatory models of CFS are based on a too narrow focus on the symptoms of CFS. In this way, the emphasis is taken away from adolescents pre-reflective, bodily and intersubjective being in the world. It was argued that underlying the overwhelming experience of fatigue and functional impairment there is a profound alteration of adolescents' relational, emotional and bodily self-experience.

SAMENVATTING

Eén van de meest kenmerkende aspecten van het chronisch vermoeidheidssyndroom (CVS) is het aanhoudende gebrek aan consistente bevindingen van ofwel 'fysieke', ofwel 'psychische' pathologie. Terwijl er een continue zoektocht is naar mogelijke verklaringen van de ernstige vermoeidheid en functionele beperking, is er weinig aandacht voor verdere verheldering van het fenomeen CVS op zichzelf. De vraag; "Wat proberen we -op ervaringsniveau- in wezen eigenlijk te verklaren?", lijkt nauwelijks gesteld te worden. Het eerste doel van deze dissertatie was dan ook te komen tot een beter begrip van de zelf-ervaring van adolescente patiënten met het syndroom. Het tweede doel was te onderzoeken of begeleiding van deze patiënten gebaseerd op hun zelf-ervaring zou leiden tot een verbetering van hun gezondheid.

Hoofdstuk 1 beschrijft de achtergrond van het in deze dissertatie beschreven onderzoek. CVS is altijd het onderwerp geweest van verhitte discussies tussen patiënten, behandelaars en wetenschappelijke onderzoekers. Een belangrijke oorsprong van deze controverses is gelegen in de veronderstelling dat patiënten met CVS op één of andere manier 'anders' zijn; ze zouden een bepaald type persoon zijn, en hun 'subjectieve' ervaring van zichzelf zou verschillen van die van gezonde individuen en van die van patiënten met een duidelijke lichamelijke aandoening. Daarom lijkt een analyse van de beschikbare persoonlijkheidsstudies in CVS een logisch startpunt. Het belangrijkste doel van dit hoofdstuk was dan ook een overzicht te geven van het voorhanden zijnde wetenschappelijk onderzoek naar persoonlijkheid en CVS. In deze studies werden er enige aanwijzingen gevonden dat de persoonlijkheden van patiënten met het syndroom verschilden van die van gezonde personen. Echter, wanneer de persoonlijkheden van patiënten met CVS werden vergeleken met andere patiënten met een chronische aandoening waren de verschillen veel minder duidelijk. Ook m.b.t. persoonlijkheid en CVS lijken er eigenlijk weinig consistente bevindingen te zijn. Daarom werd dan ook de hypothese naar voren gebracht dat meer inzicht in CVS verkregen zou kunnen worden door de zelf-ervaring, de zelfpositionering en de verhalen die patiënten met het syndroom over zichzelf vertellen te bestuderen. Voor het design van zo'n onderzoek werd een longitudinale opzet, het strikte gebruik van de CDC-criteria voor CVS, en de inclusie van gezonde individuen van

hetzelfde geslacht en met dezelfde leeftijd en patiënten met een somatische, chronische ziekte als controlegroepen aangeraden.

Hoofdstuk 2 beschrijft sommige van de uitgangskarakteristieken van de deelnemers die in de hoofdstudie van deze dissertatie geïncludeerd werden. Daarnaast worden kort de theoretische hoofdaannames m.b.t. het zelf en zelfervaring geïntroduceerd. In dialogisch zelf theorie wordt identiteit begrepen als voortkomend uit verschillende zelfposities. Volgens deze theorie wordt het zelf niet alleen geconstrueerd door dialogen met andere personen, maar ook door dialogen tussen (en over) verschillende perspectieven binnen het zelf. In het hoofdstuk worden de resultaten m.b.t. de zelfpositionering van 42 adolescenten met CVS, 37 adolescenten met jeugdreuma en 23 gezonde jongeren gepresenteerd. De patiënten met CVS rapporteerden ernstige fysieke en psychosociale problemen. Zij presenteerden zich significant minder sterk en meer onwel in hun sociale relaties dan gezonde jongeren en adolescenten met jeugdreuma. Er was een sterke indicatie dat bij jongeren met CVS posities m.b.t. ziekte buitengewoon dominant waren.

Hoofdstuk 3 beschrijft een verdere uitdieping van deze bevindingen. Het hoofdstuk beschrijft de zelfverhalen die aan de bovengenoemde zelfpositionering ten grondslag liggen. Daarnaast wordt het belangrijkste onderzoeksinstrument van de studie -de zelfkonfrontatiemethode (ZKM)- geïntroduceerd. De ZKM is een methode om de verhalen die personen over zichzelf vertellen te onderzoeken en eventueel te veranderen. Het combineert daarbij een focus op verbale uitingen met een analyse van de onderliggende emotionele en motivationele structuren. Adolescenten met CVS rapporteerden significant minder positieve ervaringen van autonomie en succes in vergelijking met gezonde jongeren. Daarnaast beschreven zij significant meer negatieve ervaringen van machteloosheid, isolement en onvervuld verlangen in vergelijking met niet alleen gezonde adolescenten, maar ook in vergelijking met jongeren met jeugdreuma. Het werd daarom van het grootste belang geacht de zelfpositionering en zelfverhalen van patiënten met CVS in een klinische setting centraal te stellen.

Hoofdstuk 4 beschrijft de resultaten van een afzonderlijke studie naar de prognose van CVS in de adolescentie na conventionele zorg -daar de hoofdstudie geen wachtlijst controleconditie had. Een cohort van 60 jongeren met CVS, die eerder in het Universitair Medisch Centrum Utrecht gezien

waren, werd na gemiddeld 2,2 jaar vervolgd. Ongeveer de helft van de patiënten leek na deze tijd hersteld. De andere helft was echter nog steeds ernstig vermoeid en fysiek beperkt. Ondanks een intensief gebruik van de gezondheidszorg bleek een substantieel deel van de jongeren met CVS dus gedurende lange tijd symptomatisch te blijven. Een slecht behandelingsresultaat was geassocieerd met een hoge mate van lichamelijke pijn, en een lage zelfwaardering, mentale- en algemene gezondheid. Dit werd gezien als een verdere indicatie dat verbetering van de bestaande behandelingsmogelijkheden en onderzoek naar additionele interventies noodzakelijk waren.

Hoofdstuk 5 beschrijft de resultaten van de hoofdstudie naar de begeleiding van de patiënten met CVS d.m.v. zelfonderzoek, en de longitudinale veranderingen in hun zelf-ervaring. De adolescente patiënten met CVS, beschreven in het tweede en derde hoofdstuk, werden gerandomiseerd en kregen vervolgens 6 dan wel 12 sessies met de ZKM toegewezen. De gezonde jongeren kregen allen 6 sessies. Zelfonderzoek leidde tot significante veranderingen in de zelfverhalen van de deelnemers. Bij de jongeren met CVS bleek er tevens een significante verbetering te zijn in hun vermoeidheid, en hun fysieke- en psychosociale functioneren. De patiënten die 12 sessies toegewezen hadden gekregen verbeterden het meest. Tien maanden na afloop van de begeleiding was er geen verslechtering opgetreden in de toestand van de patiënten. Er werd dan ook geconcludeerd dat zelfonderzoek niet alleen leidt tot gezondheidsverbetering, maar ook tot een verandering van de zelf-ervaring van adolescente patiënten met CVS. De ZKM maakt het voor deze jongeren mogelijk om zelf factoren te ontdekken die bijdragen aan hun vermoeidheid, of deze zelfs veroorzaakt. De resultaten wijzen erop dat zelfonderzoek een nuttig instrument is in de behandeling van CVS tijdens de adolescentie.

Hoofdstuk 6 is de afsluitende algemene discussie. In dit hoofdstuk wordt een eerste poging ondernomen een nieuw conceptueel kader te schetsen om de zelf-ervaring in CVS gedurende de adolescentie te begrijpen. Het start met de aanname dat de heersende modellen van het syndroom gebaseerd zijn op een te eenzijdige focus op de symptomen van CVS. In het hoofdstuk wordt beargumenteerd dat er aan de symptomen van vermoeidheid en functionele beperkingen een overweldigende verandering van de lichamelijke, emotionele en intersubjectieve zelf-ervaring van de jongeren met CVS ten grondslag ligt.

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CURRICULUM VITAE

Stefan van Geelen werd op 3 juli 1975 geboren in Amersfoort. In 1994 behaalde hij zijn VWO diploma op De Amersfoortse Berg te Amersfoort. Datzelfde jaar begon hij met de studie Wijsbegeerte aan de Universiteit Utrecht. Gedurende deze studie werkte hij als student-assistent bij de secties Ethiek, Wijsgerige Antropologie en Geschiedenis van de Filosofie van de Oudheid. In 1998 werd hij geselecteerd voor het Kyoto University International Exchange Program. Tot en met het voorjaar van 1999 woonde en studeerde hij in Kyoto, Japan. Hij volgde daar zijn niet-wijsgerig bijvak Japanse Taal & Cultuur. Begin 2001 studeerde hij af in de Sociale & Politieke Filosofie. Aansluitend werkte hij korte tijd als junioronderzoeker bij de vakgroep Praktische Filosofie van de Faculteit Wijsbegeerte van de Universiteit Utrecht. Eind 2001 begon hij bij de Divisie Kinderen van het Universitair Medisch Centrum Utrecht als junioronderzoeker met een pilotstudie naar de belevingswereld van jongeren met het chronisch vermoeidheidssyndroom. In 2004 begon hij bij dezelfde divisie met het onderzoek dat tot dit proefschrift leidde. Gedurende het wintersemester 2008/ 2009 werkte hij als gastonderzoeker bij de sectie Fenomenologische Psychopathologie van het Departement Psychiatrie van de Universiteit van Heidelberg, Duitsland. In 2009 begon hij in het kader van de 'Skills Labs', samen met collega's, met de ontwikkeling en uitvoering van een weerbaarheidstraining voor chronisch zieke kinderen. Daarnaast werkt hij momenteel, in samenwerking met het Ethiek Instituut van de Faculteit Geesteswetenschappen, aan een project over de morele aspecten van bariatrische chirurgie bij kinderen en adolescenten met morbide obesitas.

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