

A population-based study of chronic fatigue syndrome (CFS) experienced in differing patient groups: An effort to replicate Vercoulen et al.'s model of CFS

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Abstract

Background: Vercoulen et al.'s (1998) model characterizes patients with chronic fatigue syndrome (CFS) as having insufficient motivation for physical activity or recovery, lacking an internal locus of control, and maintaining a self-defeating preoccupation with symptoms. However, this model has only been tested in a poorly specified group using a single comparison sample.

Aims: To investigate whether Vercoulen et al.'s model provides an adequate description of CFS in a community-based sample.

Method: A community-based sample recruited through telephone interviewing ($N = 28,763$) produced five groups (CFS, CF-psychiatrically-explained symptoms, CF-medically-unexplained symptoms, CF-substance misuse, and idiopathic CF). The data were analysed using path analysis with the endogenous (dependent) variables, fatigue severity, physical activity, and impairment, were ratio-level measurements and consisted of at least four values. The exogenous (independent) variables except for causal attribution of fatigue were also ratio-level measurements.

Results: The current investigation found that the Vercoulen et al. model adequately represented chronic fatigue secondary to psychiatric conditions but not CFS.

Conclusions: This finding points to important differences between CFS and psychiatrically-explained chronic fatigue which may have an impact on the development of therapy as well as explanatory models.

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An effort to replicate Vercoulen et al.'s Model of CFS

Fatigue is a significant problem for about 25% of all patients seen by general practitioners (Chen, 1986; Kroenke, Wood, Mangelsdorff, Meier, & Powell, 1988). Approximately 5% of individuals have what is known as chronic fatigue, which involves unexplained fatigue that persists for six or more months (Price, North, Wessely, & Fraser, 1992; Pawlikowska et al., 1994; Jason et al., 1995). Some individuals with chronic fatigue have an illness called chronic fatigue syndrome (CFS) (Mechanic, 1993), and while several biological markers have been suggested for this illness, none occur for all patients with CFS (Friedberg & Jason, 1998; Taylor, Friedberg, & Jason, 2001).

In part, due to the lack of a biologic marker, several theorists have proposed psychogenic approaches for understanding CFS (Wessely, Hotopf & Sharpe, 1998).

For some, CFS was assumed to be a psychologically-determined problem (Manu, Lane, & Matthews, 1988). These views ultimately affected some physicians who believed that CFS was similar to neurasthenia, and that CFS would eventually have a similar fate once people recognized that most patients with this disease were really suffering from a psychiatric illness. Complicating this situation was the fact that psychiatrists and physicians have also regarded fatigue as one of the least important of presenting symptoms (Lewis & Wessely, 1992).

These biases have been filtered to the media, which has portrayed CFS in simplistic and stereotypic ways. For example, after the Sharpe et al. (1996) treatment study was published, British tabloids came out with headlines such as: “ME’s (the term for CFS in England) Mainly in the Mind: Study reveals yuppie flu can be cured by positive thinking”. One major consequence is that many CFS patients feel dissatisfied with their medical care (David, Wessely, & Pelosi, 1991) and have gone outside traditional medicine to be treated for their illness (Denz-Penhey & Murdoch, 1993; Jason, Ferrari, Taylor, Slavich, & Stenzel, 1996).

As an example of this psychogenic approach within the CFS literature, Vercoulen et al. (1998) developed a model using a sample of 50 patients with CFS and a sample of 51 individuals with multiple sclerosis. They concluded that while both groups attributed their symptoms to physical causes, only the group with CFS demonstrated a significant association between somatic attributions and decreased physical activity, and decreased physical activity and increased fatigue severity. Vercoulen et al. characterized individuals who suffer from CFS as inclined to improperly associate physical activity with a worsening of symptoms. Figure 1 provides a diagram of the Vercoulen et al. model. As this is one of the few studies to actually evaluate a model, the paper has received considerable attention among CFS scholars. However, while it has been cited frequently, no critical reviews or replications of this study have been published.

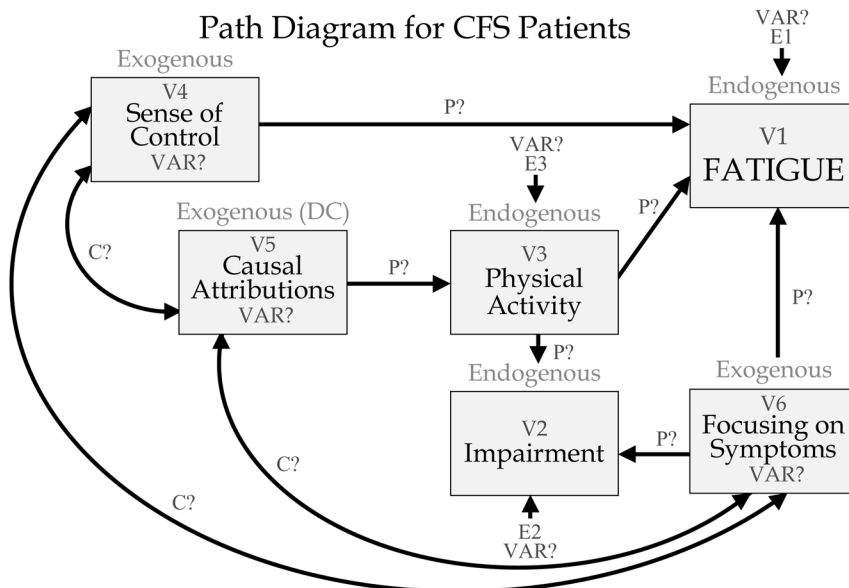


Figure 1. Path diagram of the Vercoulen et al. model.

Vercoulen et al. (1998) tested their model using structural equation modeling, but there were several possible limitations in this study, as the authors: (1) did not distinguish latent and observed variables; (2) did not analyze the structural and measurement model as separate components; (3) did not achieve proper model identification based on the relationship between their sample data covariance matrix and the theoretical model population covariance matrix; and (4) did not discuss distributive properties of their variables (multivariate normality, skew, etc.).¹ A final possible problem in the Vercoulen et al. (1998) study is that these investigators did not indicate whether the sample met criteria for CFS based on the Fukuda et al. (1994) case definition, which are the criteria used by most investigators. It is possible that some participants with CFS in the Vercoulen et al. (1998) study might not have met the Fukuda et al. research case definition.

Given the statistical and diagnostic issues reviewed above, it was important to attempt to replicate the Vercoulen et al. (1998) model with a more clearly diagnosed sample. In the present study, several distinct groups of participants were studied; those diagnosed with CFS, those diagnosed with chronic fatigue due to psychiatric explanations, those with medical reasons for their chronic fatigue, those diagnosed with substance abuse as a reason for their fatigue, those with Idiopathic chronic fatigue, and controls. In this study, we evaluated the fit of Vercoulen et al.'s model for each of these groups.

Methods

The data derived from a multi-stage community-based study examining the prevalence of chronic fatigue syndrome in a socioculturally diverse urban sample. Stage 1 entailed a cross-sectional screening telephone survey of a random sample of 28,673 households. Stage 2 involved a structured psychiatric interview of those respondents from Stage 1 who screened positive for CFS-like illness and of a random sample of individuals who tested negative. Stage 3 was comprised of a medical exam and structured history taking of the experimental and control groups. Stage 4 entailed the examination of psychosocial measurements.

Procedure

Stage 1. A random sample of adults 18 years or older, selected by the most recent birth date in each contacted household, was screened during a period of twenty months. Of the 28,673 households telephoned, completed interviews occurred for 18,675 households (65.1% completion rate). One adult was interviewed in each household. The sample was stratified to represent the neighborhoods of Chicago, Illinois that were 10 to 15 minutes from the site of the medical examination. Eight Chicago area communities were sampled including the low SES area of West Garfield Park, middle-income areas of Bridgeport and Armour Park, gentrified area of the near West Side, and high SES area of the Loop and near North Side. Sample Survey Incorporated randomly generated listed and unlisted telephone numbers that had valid Chicago prefixes (See Jason et al., 1999a and 1999b for more details).

There were 780 individuals who screened positive for chronic fatigue. Of these individuals with 6 or more months of fatigue, 304 participants (39%) did not endorse the minimum number of symptoms necessary to qualify for a CFS diagnosis, 68 participants (8.7%) had chronic fatigue that was associated with either clinical mood impairment, an eating or psychotic disorder, substance abuse, or a medical condition, and 408 participants (52.3%) presented with CFS-like profiles (Fukuda et al., 1994).

Stage 2. All 408 members of the CFS-like group were invited to participate in the next phase of the study that entailed the completion of the psychiatric assessment. In addition, we randomly selected a group of 199 screened negative controls. The control group was composed of individuals selected randomly from those 18,260 screened negatives (groups included participants with no prolonged or chronic fatigue, prolonged fatigue, ICF-like illness, and CF-explained-like illness). This group was composed primarily of individuals with no profound or chronic fatigue, as 88.1% of the sample consisted of such individuals.

Stage 3. Medical assessments occurred on 166 patients with CFS-like illness and 47 controls, and these are the individuals who agreed to come in for a medical appointment. There were no significant differences between the 166 screened positive (CFS-like) participants and the 242 screened positive (CFS-like) non-participants on fatigue scale scores, gender, ethnic identification, age, occupation, education, and marital status. Identical analyses were executed to compare participants and non-participants selected within the screened negative group (e.g., individuals in the no fatigue, prolonged fatigue, ICF-like, and CF-explained-like groups). Analogous to findings within the screened positive group, analysis comparing similar fatigue and demographic indices between the 47 screened negative participants and the 152 screened negative non-participants revealed no significant differences between groups.

Stage 4. Psychosocial measures were administered to the participants.

Sample. The physician review team evaluated the data of the five diagnoses generated by the physician reviews were chronic fatigue syndrome (CFS), psychiatrically-explained (CF-psychiatric), medically-explained chronic fatigue (CF-medical), idiopathic chronic fatigue (ICF), and chronic fatigue related to substance abuse (CF-substance related). These categories are summarized in Table I. Those with psychiatric reasons for their fatigue had psychiatric conditions that were exclusionary for a CFS diagnosis (e.g., melancholic depression) based on the Fukuda et al. (1994) criteria. Those with medical reasons for their fatigue had medical conditions that were exclusionary for a CFS diagnosis (e.g., cancer) based on the Fukuda et al. (1994) criteria. Those with ICF did not have the necessary number of somatic symptoms for a CFS diagnosis. Finally, those with substance abuse within two years of onset of CFS were exclusionary of a CFS diagnosis because of the Fukuda et al. criteria.

Table I. Chronic fatigue groupings.

Fatigue category	Number of participants
Total selected for Phase 2 Evaluation	408
Total completed Physician Review Phase	166
Final diagnosis	
Chronic Fatigue Syndrome	32
CF – psychiatrically explained	33
CF – medically explained	33
CF – explained by substance abuse	23
Idiopathic chronic fatigue	45

Measures

The Medical Outcomes Study 36-item Short Form (MOS). The Medical Outcomes Study 36-item Short Form Survey (MOS) (Ware & Sherbourne, 1992; Ware, Snow, Kosinski & Gandek, 2000), a reliable and valid measure that discriminated between gradations of disability. The instrument encompassed multi-item scales that assessed physical functioning, role limitations, social functioning, bodily pain, general mental health, vitality, and general health perceptions. Higher scores indicated better health, lower disability, or less impact of health on functioning. Reliability and validity studies for the 36-item version of the MOS have shown adequate internal consistency, discriminant validity among subscales, and substantial differences between patient and non-patient populations in the patterns of scores (McHorney, Ware, Rogers, Raczek & Lu, 1992; McHorney, Ware & Raczek, 1993; McHorney, Ware, Lu & Sherbourne, 1994). This was the measure used in the present study for assessing degree of impairment.

The Perceived Stress Scale. The Perceived Stress Scale (Cohen, Kamark, & Mermelstein, 1983) is a four-item revised version of a previous fourteen-item measure of global perceived stress. The time period tapped into by this instrument was the last month. The authors report a coefficient alpha reliability of .72 for the four-item short version. Total stress scores range from 0–16 with higher scores measuring greater stress.

The Illness Management Questionnaire (IMQ). The Illness Management Questionnaire (IMQ) was developed specifically to assess coping in patients with CFS and has been used in studies of adults with CFS (e.g., Ray, Jefferies, & Weir, 1995). The IMQ produced scores on four different factors: Maintaining Activity (attempting to ignore symptoms, disregarding possible adverse effects of activity), Accommodating to the Illness (organizing and arranging one's life to avoid exertion and manage stress), Focusing on Symptoms (preoccupation with symptoms, viewing one's life as dominated by the illness) and Information Seeking (searching for relevant information and an openness to try treatments). The instrument provides a measure of problem-focused coping. Similar to the Perceived Stress Scale, the time period tapped into by this instrument was the past month. Test-retest reliabilities of the IMQ ranged from .85 (Information Seeking) to .93 (Maintaining Activity). Higher scores on the IMQ indicated greater use of the coping style. The Scale that was used in the present study was Focusing on Symptoms.

Activities Questionnaire. The Activities Questionnaire was used to rate participants' physical activity level in terms of their social/recreational and vocational functioning. The physical activity variable in the current study was based on participants' self-assessment of their social and vocational activity levels during their most impaired periods. Ratings were based on a ten-point Likert scale where 0 reflected total absence of physical activity and 10 reflected no reduction in pre-illness physical activity.

Causal Attributions. Participants were asked what was causing their fatigue and to characterize their attributions as having physical or psychological origins. The causal attribution question required respondents to choose one of five explanations, "definitely physical", "mainly physical", "equally physical and psychological", "mainly psychological", and "definitely psychological" causes. We categorized answers to this question into three discrete causal attribution cut points, 1 = physical causes, encompassing the definitely physical and mainly physical causal attribution groups; 2 = equally physical and psychosocial

causes; and 3 = psychosocial causes, encompassing the definitely psychological and mainly psychological causal attribution groups.

Fatigue Scale. The Fatigue Scale (Wessely & Powell, 1989) is a well regarded measure tapping the construct fatigue, and it has been used in a community-based study of fatigue in Great Britain (Pawlikowska et al., 1994). This scale produces a total fatigue score, a score reflecting mental fatigue, and a score reflecting physical fatigue. Chalder et al. (1993) further refined the Fatigue Scale; and despite its brevity, the scale has been found to be reliable and valid. It has good face validity and reasonable discriminant validity. This 11-item measure has as possible responses “less than usual”, “no more than usual”, “more than usual”, and “much more than usual”. Total fatigue scores, which were the indicator used in the present study, ranged from 0–33 with higher scores signifying greater fatigue.

Statistical analyses

In order to replicate the Vercoulen et al. model (Figure 1), we assessed their model with our CFS, CF-psychiatric, CF-medical, CF-substance related, ICF and control groups. In other words, the Vercoulen et al. model was tested six times, once with each of these six groups.

The analyses in the current investigation were based on Hatcher's (1994) approach to path analysis with manifest variables using the SAS system. The assumptions and conditions for testing a structural equation model (Hatcher, 1994) were satisfied. The endogenous (dependent) variables, fatigue severity, physical activity, and impairment, were ratio-level measurements and consisted of at least four values. The exogenous (independent) variables except for causal attribution of fatigue were also ratio-level measurements, and since causal attribution of fatigue was a classification variable its three levels were dummy-coded. Various SAS-based tests of multivariate kurtosis suggested that multivariate normality of the data was a reasonable assumption. Additionally, the variables in the model were free of multicollinearity since there were not any correlations stronger than $r = -.49$, which was the coefficient for the association between focusing on symptoms and physical activity in the CFS group. The models were over-identified, so tests of goodness of fit could be considered accurate and reliable. The number of data points, 21, exceeded the number of parameters to be estimated, 15, which was necessary for model identification. Convergence was achieved in five to 17 iterations depending on the combination of chronic fatigue subgroup and causal attribution level being tested, and the convergence criteria were satisfied for all the models.

Below we describe the goodness of fit tests generated in the SAS output that were used to evaluate whether the groups fit the Vercoulen et al. model. The criteria included the normalized residual matrix, chi-square (χ^2) goodness-of-fit index, non-normed fit index (NNFI), comparative fit index (CFI), and R^2 .²

A power analysis for covariance structure modeling (MacCallum, Browne, & Sugawara, 1996) indicated that adequate sample size to achieve moderate statistical power, $\beta_M = 0.50$, was $n_M = 242$. In this computation, there were $p = 6$ manifest variables, $q = 3$ covariance parameters, and $df = 18$ degrees of freedom. Clearly, power was a concern in this study, but if the model successfully described one or more of the groups, then that would be an indication that had the sample size been larger, more robust findings would have emerged.

Results

The Vercoulen et al. model demonstrated adequate fit for the CF-psychiatric sample only. The covariance matrices for the CF-psychiatric group are in Table II. There is one matrix

Table II. Covariance matrices for the CF-psychiatric sample.

Focus CF-Psychiatric	Fatigue	Impairment	Phys. Activ.	Sense control	Phys. cause	symptoms
Causal attribution of fatigue – physical						
Fatigue	1.0000.					
Impairment	– 0.0963	1.0000				
Phys. activity	– 0.3241	– 0.3737	1.0000.			
Sense control	0.1491	– 0.2780	0.2754	1.0000		
Physical cause	0.0557	0.0203	– 0.2852	– 0.1684	1.0000.	
Focus symptoms	0.1049	– 0.3019	0.0841	0.3845	0.3683	1.0000
Causal attribution of fatigue – equally physical and psychosocial						
Fatigue	1.0000					
Impairment	– 0.0963	1.0000				
Phys. activity	– 0.3241	– 0.3737	1.0000			
Sense control	0.1491	– 0.2780	0.2754	1.0000		
Phys. psych.	0.2967	– 0.1325	0.0656	0.2835	1.0000.	
Focus symptoms	0.1049	– 0.3019	0.0841	0.3845	– 0.1657	1.000
Causal attribution of fatigue – psychosocial						
Fatigue	1.0000					
Impairment	– 0.0963	1.0000				
Phys. activity	– 0.3241	– 0.3737	1.0000			
Sense control	0.1491	– 0.2780	0.2754	1.0000		
Psych. cause	– 0.1671	0.1812	0.0611	– 0.0421	1.0000.	
Focus symptoms	0.1049	– 0.3019	0.0841	0.3845	– 0.1237	1.0000

for each causal attribution level. These values were necessary to compute the fit criteria that determined the degree of fit of the CF-psychiatric group to the Vercoulen et al. model. For the CF-psychiatric group, the χ^2 statistic was associated with a non-significant p -value ($> .05$), the CFI and NNFI were greater than .9, there were no elements in the residual matrix with an absolute value larger than |2.00|, and the R^2 value was adequate at each causal attribution level of fatigue (See Table III).

The Vercoulen et al. model did not produce adequate fit indices for the CFS, CF-medical, ICF, CF-substance related or healthy control groups, at any of the causal attribution levels, physical, equally physical and psychosocial, and psychosocial. Table III provides a summary of the fit indices for each individual chronic fatigue group. The values in italics indicate insufficient model fit. For at least one causal attribution level within these other fatigue groups, the χ^2 statistics were associated with significant p -values ($< .05$), the CFI's and NNFI's were less than .9, there were elements in the residual matrices with an absolute value greater than |2.00|, and the R^2 values were inadequate.

An additional analysis was conducted on the combined CFS and CF-medical groups to evaluate whether increasing the sample size would lead to better model fit. Combining the samples still did not produce adequate fit indices. Table IV provides a summary of the fit statistics. At all causal attribution levels, the CFI, NNFI and R^2 indicated inadequate model fit. In separate analyses, combining either the CFS, CF-medical and CF-psychiatric groups or combining all six groups continued to produce poor model fit between the sample data and the Vercoulen et al. model, which further demonstrated that increasing sample size alone did not enhance the fit of the Vercoulen et al. model.

Table III. Summary of Vercoulen et al. (1998) model replication.

Group/Attribution	χ^2	CFI	NNFI	R ²	Residuals
<i>CFS</i>					
Physical	0.001	0.019	- 1.452	33.0%	6/10 > 2.00
Phys. & Psych.	0.004	0.170	- 1.076	31.5%	5/10 > 2.00
Psychosocial	0.028	0.314	- 0.715	31.5%	5/10 > 2.00
<i>CF-Psychiatric</i>					
Physical	0.578	1.000	1.327	43.0%	0/10 > 2.00
Phys. & Psych.	0.477	1.000	1.138	41.0%	0/10 > 2.00
Psychosocial	0.653	1.000	2.498	41.5%	0/10 > 2.00
<i>CF-Medical</i>					
Physical	0.951	None	- 0.554	26.5%	0/10 > 2.00
Phys. & Psych.	0.978	None	- 0.608	25.0%	0/10 > 2.00
Psychosocial	None	None	None	None	None
<i>CF-Substance abuse</i>					
Physical	None	None	None	None	None
Phys. & Psych.	0.231	0.854	0.636	60.0%	0/10 > 2.00
Psychosocial	0.289	0.900	0.751	59.0%	0/10 > 2.00
<i>ICF</i>					
Physical	0.089	0.914	0.785	59.0%	0/10 > 2.00
Phys. & Psych.	0.198	0.922	0.806	69.0%	0/10 > 2.00
Psychosocial	0.305	0.956	0.890	59.0%	0/10 > 2.00
<i>Healthy controls</i>					
Physical	0.018	0.300	- 0.751	19.0%	5/10 > 2.00
Phys. & Psych.	0.025	0.428	- 0.431	25.3%	0/10 > 2.00
Psychosocial	None	None	None	None	None

The values in italics indicate insufficient model fit.

Table IV. Model replication with CFS and CF-medical groups.

Group/Attribution	χ^2	CFI	NNFI	R ²	Residuals
<i>CFS & CF-medical</i>					
Physical	> 0.05	0.41	- 0.47	19%–21%	0/10 > 2.00
Phys. & Psych.	> 0.05	0.78	0.45	19%–21%	0/10 > 2.00
Psychosocial	> 0.05	0.70	0.25	19%–21%	1/10 > 2.00

The values in italics indicate insufficient model fit.

Discussion

The aim of this study was to replicate the Vercoulen et al. model with a more clearly diagnosed sample and with a clearer data analytic description of the structural equation modeling. The sample consisted of five fatigue-related diagnostic groups, and one control group, with the number in each group ranging from 23 to 45 participants. This study was only able to replicate the model for participants with psychiatric conditions, and not CFS. Vercoulen et al. (1998) suggested their findings indicated that individuals with CFS attribute their symptoms to physical causes, are overly preoccupied by physical limitations, and do not maintain regular activity. According to this model, these factors cause individuals with CFS to be functionally impaired and to experience severe fatigue. The

fact that this model could not be replicated with either the CFS or those with medical reasons for their chronic fatigue, those diagnosed with substance abuse as a reason for their fatigue, those with idiopathic chronic fatigue, and controls suggests an important distinction between individuals with chronic fatigue due to a psychiatric condition versus CFS. In other words, the present study does not support a purely psychogenic explanation for CFS.

There are several possible reasons for the discrepancies between the findings of the present study and that of Vercoulen et al. The present study was able to evaluate individuals who had been diagnosed with CFS, whereas the participants used in the Vercoulen et al. study might not have met the Fukuda et al. criteria. It is at least possible that patients with CFS from the Vercoulen et al. study were more similar to the present study's CF-psychiatric group. It is also unclear whether proper model identification was achieved in the Vercoulen et al. study and it is difficult to evaluate the parameter estimation method that was used, as those authors did not clearly discuss their computational methods. Finally, differences between recruiting patients from tertiary care settings versus community-based samples might also explain some of the differences.

Vercoulen et al.'s model clearly emphasizes psychogenic, cognitive and behavioral factors in the etiology and maintenance of CFS. In a sense, Vercoulen et al.'s model has many similarities to what has been referred to as functional somatic syndrome, which is characterized by diffuse poorly defined symptoms that cause significant subjective distress and disability, and cannot be corroborated by consistent documentation of organic pathology. Some theorists have even argued that syndromes such as CFS, Fibromyalgia (FMS) and Irritable Bowel Syndrome (IBS) may be better understood in terms of a unitary model of functional somatic distress, rather than as separate diagnostic entities (Barsky & Borus, 1999).

A study by Taylor, Jason and Schoeny (2001) evaluated the diagnostic validity of conditions that have been labeled functional somatic syndromes. Latent variable models of functional somatic distress were estimated from medical questionnaire items that closely conformed to formal diagnostic criteria for the conditions were used in model estimation. Results of confirmatory factor analysis supported diagnostic distinctions between five syndromes (Fibromyalgia, CFS, somatic depression, somatic anxiety, and irritable bowel syndrome). Discrete diagnostic categories of Fibromyalgia and CFS were then tested using logistic regression analysis, in which the outcome involved independent diagnosis of these conditions based upon physician evaluation. The diagnostic validity of the latent constructs of FMS and CFS emerging from this five-factor model were further cross-validated using findings from an independent physician evaluation.

Additional support of findings for distinctions among these syndrome constructs comes from the work of Hickie, Koschera and associates (1999), who found that chronic fatigue is a persistent diagnosis over time, and that longitudinal patterns of comorbidity of fatigue with psychological distress did not suggest a causal relationship or common vulnerability factor. Similar findings of a study by Van Der Linden and associates (1999) supported the existence of a pure independent fatigue state over time where this pure fatigue state did not predict subsequent psychiatric disorder. Morriss and associates (1999) also found that depression was not associated with the reporting of pain, FMS, IBS, and medically-unexplained symptoms in individuals with CFS.

Recent studies by the group of Dutch investigators that developed the Vercoulen model raise additional questions about their model. As an example, when 20 ambulant patients with CFS were compared to controls, there were no differences among indices of physical fitness (Bazelmans, Bleijenbergh, van der Meer, & Folgering, 2001); therefore,

this study suggested that deconditioning is not a perpetuating factor in CFS. Werf et al. (2000) found no relationship between type of activity pattern among patients with CFS and levels of depressive symptoms. In addition, approximately one fourth of a sample of CFS patients differed significantly from control patients in that they were pervasively passive. Werf et al. concluded that a proportion of patients with CFS have activity patterns that are comparable to those of controls, and therefore activity regulation might be more important for these active CFS patients, whereas the passively active group might benefit more from increasing physical activity. Finally, in a recent dissertation, Werf et al. (2003) found a relationship between daytime fatigue and physical activity in only 12% of the CFS sample, and they suggest that this indicates little support for the activity-fatigue association that has been reported by patients. Patients with CFS did report more fatigue than controls at each time point, but the CFS group did not differ with respect to their evening and night-time physical activity scores. Pervasively active patients did report an increase in fatigue during the day compared to the less active patients. This sample of pervasively active patients therefore resembled more closely the hypothesized exertion-related fatigue, but this is the very group that is least likely to be deconditioned. In contrast to Werf et al.'s explanation, it is possible that the lack of relationship between activity and fatigue among most patients is that they have learned to modify their activities to avoid fatigue, and that the relationship between fatigue and activity only occurs for those who are most active, which are those that are still pushing themselves beyond their energy envelopes.

Rather than conceptualizing CFS as a disease of solely psychiatric or behavioral factors (as is suggested by the study by Vercoulen et al., 1998), a biopsychosocial perspective suggests that complex interactions between multiple biological and psychological factors influence the onset of CFS and pathways to further illness or recovery. The biopsychosocial model (Friedberg & Jason, 1998) contends that there might be multiple pathways leading to the cause and maintenance of the neurobiologic dysregulations and other symptoms experienced by individuals with CFS. Depending upon the individual, these may include unique biological, genetic, neurological, psychological, and socioenvironmental contributions. Attempts to understand CFS need to take into account the multiple losses in jobs, diminished relationships, financial instability, and disruption in future planning, daily activity, extracurricular interests, stamina and spontaneity (Anderson & Ferrans, 1997). To better understand CFS, the effects of these types of pervasive losses need to be examined, as the stigma associated with CFS affects coping and health outcomes (Friedberg & Jason, 1998).

To the extent that a purely psychogenic explanation is embraced by the research community, important more medically-oriented research attempting to understand the pathophysiology of this illness might be avoided. In addition, a purely psychogenic explanation might cause health care workers to focus all treatment efforts on non-pharmacologic approaches, rather than a healthy mixture of both pharmacologic and non-pharmacologic treatments.

There were several limitations in the present study. Several of the measures used were designed specifically for this study, and there were no reliability estimates for them. Another important limitation of the present study was the small sample sizes. However, the fact that the Vercoulen et al. model did fit with the chronic fatigue group due to psychiatric reasons does indicate that there was enough power in even this small sample. Clearly, there is a need for additional studies in this area, with larger sample sizes. Such studies are needed so that we can begin to develop models that acknowledge biological and contextual elements and that might better capture the CFS illness.

Notes

- 1 In Vercoulen et al.'s (1998) model, they did not distinguish latent and observed variables in their measurement model. Latent variables are not directly measurable; therefore, they generate measurement error that needs to be accounted for (Schumacker & Lomax, 1996). Additionally, the measurement model provides an assessment of both convergent and discriminant validity whereas the structural model provides an assessment of nomological validity. As unique information is contained in each component of the structural equation model, it is unclear why Vercoulen et al. did not analyze the measurement model and the structural model as separate components. It is also unclear whether proper model identification was achieved as it is ambiguous whether Vercoulen et al. established a unique set of parameter estimates based on the relationship of the sample data covariance matrix to the theoretical model population covariance matrix. In addition, neither the estimation procedures nor multivariate normality, skew or kurtosis were discussed, therefore, it is difficult to evaluate the parameter estimation method that the researchers used. In regard to the criteria with which the authors evaluated each model stage, they assessed fit on the basis of two criteria, by obtaining a significant χ^2 and by maximizing the adjusted goodness-of-fit index. Vercoulen et al. could have reduced specification error further by including such criteria as ones for comparing models with different degrees of freedom (i.e., Normed Fit Index and Parsimonious Fit Index) and ones for comparing models with varying numbers of latent variables (i.e., Akaike Information Criterion).
- 2 Hatcher's (1994) instructions for interpreting these criteria are discussed. When conducting path analysis, if the theoretical model successfully accounts for the hypothesized relationships between the variables, then the theoretical covariance matrix will be nearly identical to the sample-based covariance matrix. Each element of the residual matrix will be zero or near zero. If there are elements in the residual matrix that have an absolute value larger than 2.00, the theoretical model likely contains specification errors. The χ^2 statistic examines the null hypothesis that the theoretical model fits the data. Since rejection of the alternate hypothesis that the model does not fit the data is desired, the obtained χ^2 value needs to be small and the p-value associated with the χ^2 needs to be greater than .05. The non-normed fit index (NNFI) supplements the chi-square goodness-of-fit test. Values greater than .9 indicate acceptable fit of the theoretical model to the data. The non-normed fit index is similar to the normed fit index criterion but the non-normed fit index better reflects model fit at all sample sizes than the normed fit index. Like the NNFI, the comparative fit index (CFI) provides accurate assessment of model fit regardless of sample size. Values of the CFI range from zero to one, and values greater than .9 indicate relatively good fit. Finally, the R^2 values indicate the percentage of variation in the endogenous variables accounted for by their direct antecedents. As in multiple regression analysis, R^2 values range from zero to one and larger values indicate a greater percentage of explained variation.

References

- Anderson, J. S., & Ferrans, C. E. (1997). The quality of life of persons with chronic fatigue syndrome. *The Journal of Nervous and Mental Disease, 185*, 359–367.
- Barsky, A. J., & Borus, J. F. (1999). Functional somatic syndromes. *Annals of Internal Medicine, 130*, 910–921.
- Bazelmans, E., Bleijenberg, G., van der Meer, J. W. M., & Folgering, H. (2001). Is physical deconditioning a perpetuating factor in chronic fatigue syndrome? A controlled study on maximal exercise performance and relations with fatigue, impairment and physical activity. *Psychological Medicine, 31*, 107–114.
- Chalder, T., Berelowitz, G., Pawlikowska, T., Watts, L., Wessely, S., Wright, D., & Wallace, E. P. (1993). Development of a fatigue scale. *Journal of Psychosomatic Medicine, 37*, 147–153.
- Chen, M. (1986). The epidemiology of self-perceived fatigue among adults. *Preventive Medicine, 15*, 74–81.
- Cohen, S., Kamarck, T., & Mermelstein, R. (1983). A global measure of perceived stress. *Journal of Health and Social Behavior, 24*, 385–396.
- David, A. S., Wessely, S., & Pelosi, A. J. (1991). Chronic fatigue syndrome: Signs of a new approach. *British Journal of Hospital Medicine, 45*, 158–163.
- Denz-Penhey, H., & Murdoch, J. C. (1993). Service delivery for people with chronic fatigue syndrome: A pilot action research study. *Family Practice, 10*, 14–18.
- Friedberg, F., & Jason, L. A. (1998). *Understanding chronic fatigue syndrome*. Washington, DC: American Psychological Association.
- Fukuda, K., Strauss, S. E., Hickie, I., Sharpe, M. C., Dobbins, J. G., & Komaroff, A. (1994). The chronic fatigue syndrome: A comprehensive approach to its definition and study. *Annals of Internal Medicine, 121*, 953–959.
- Hatcher, L. (1994). *A step-by-step approach to using the SAS system for factor analysis and structural equation modeling*. Cary, NC: SAS Institute Inc.

- Hickie, I., Koschera, A., Hadzi-Pavlovic, D., Bennett, B., & Lloyd, A. (1999). The temporal stability and comorbidity of prolonged fatigue: A longitudinal study in primary care. *Psychological Medicine*, *29*, 855–861.
- Jason, L. A., Ferrari, J. R., Taylor, R. R., Slavich, S. P., & Stenzel, C. L. (1996). A national assessment of the service, support, and housing preferences by persons with chronic fatigue syndrome: Toward a comprehensive rehabilitation program. *Evaluation and the Health Professions*, *19*, 194–207.
- Jason, L. A., Jordan, K. M., Richman, J. A., Rademaker, A. W., Huang, C., McCready, W., Shales, J., King, C. P., Landis, D., Torres, S., Haney-Davis, T., & Frankenberry, E. L. (1999a). A community-based study of prolonged fatigue and chronic fatigue. *Journal of Health Psychology*, *4*, 9–26.
- Jason, L. A., Richman, J. A., Rademaker, A. W., Jordan, K. M., Plioplys, A. V., Taylor, R. R., McCready, W., Hwang, C., & Plioplys, S. (1999b). A community based study of chronic fatigue syndrome. *Archives of Internal Medicine*, *159*, 2129–2137.
- Jason, L. A., Taylor, R. R., Wagner, L. I., Holden, J., Ferrari, J. R., Plioplys, A. V., Lipkin, D., & Papernik, M. (1995). Estimating rates of chronic fatigue syndrome from a community based sample: A pilot study. *American Journal of Community Psychology*, *23*, 557–568.
- Kroenke, K., Wood, D., Mangelsdorff, D., Meier, N., & Powell, J. (1988). Chronic fatigue in primary care: Prevalence, patient characteristics and outcome. *Journal of the American Medical Association*, *260*, 929–934.
- Lewis, G., & Wessely, S. (1992). The epidemiology of fatigue: More questions than answers. *Journal of Epidemiology and Community Health*, *46*, 92–97.
- Mac Callum, R. C., Browne, M. W., & Sugawara, H. M. (1996). Power analysis and determination of sample size for covariance structure modeling. *Psychological Methods*, *1*, 130–149.
- Manu, P., Lane, T. J., & Matthews, D. A. (1988). The frequency of the chronic fatigue syndrome in patients with symptoms of persistent fatigue. *Annals of Internal Medicine*, *109*, 554–556.
- McHorney, C. A., Ware, J. E., Lu, R. L., & Sherbourne, D. (1994). The MOS 36-item Short-Form Health Survey (SF-36): III. Tests of data quality, scaling assumptions, and reliability across diverse patient groups. *Medical Care*, *32*, 40–66.
- McHorney, C. A., Ware, J. E., & Raczek, A. E. (1993). The MOS 36-item Short-Form Health Survey (SF-36): II. Psychometric and clinical tests of validity in measuring physical and mental health constructs. *Medical Care*, *3*, 247–263.
- McHorney, C. A., Ware, J. E., Rogers, W., Raczek, A. E., & Lu, J. F. (1992). The validity and relative precision of MOS short- and long- form health status scales and Dartmouth COOP charts. Results from the Medical Outcomes Study. *Medical Care*, *30*(Suppl.), MS 253–265.
- Mechanic, D. (1993). Chronic fatigue syndrome and the treatment process. *Chronic Fatigue Syndrome Ciba Foundation Symposium*, *173*, 318–341.
- Morriss, R. K., Ahmed, M., Wearden, A. J., Mullis, R., Strickland, P., Appleby, L., Campbell, I. T., & Pearson, D. (1999). The role of depression in pain, psychophysiological syndromes, and medically unexplained symptoms associated with chronic fatigue syndrome. *Journal of Affective Disorders*, *55*, 143–148.
- Pawlikowska, T., Chalder, T., Wessely, S., Wright, D., Hirsch, S., & Wallace, P. (1994). A population based study of fatigue and psychological distress. *British Medical Journal*, *308*, 763–766.
- Price, R. K., North, C. S., Wessely, S., & Fraser, V. J. (1992). Estimating the prevalence of chronic fatigue syndrome and associated symptoms in the community. *Public Health Reports*, *107*, S14–S21.
- Ray, C., Jefferies, S., & Weir, W. R. C. (1995). Coping with chronic fatigue syndrome: Illness responses and their relationship with fatigue, functional impairment and emotional status. *Psychological Medicine*, *25*, 937–945.
- Schumacker, R. E., & Lomax, R. G. (1996). *A beginner's guide to structural equation modeling*. Mahwah, NJ: Lawrence Erlbaum Associates, Inc.
- Sharpe, M., Hawton, K., Simkin, S., Surawy, C., Hackmann, A., Klimes, I., Peto, T., Warrell, D., & Seagroatt, V. (1996). Cognitive behaviour therapy for the chronic fatigue syndrome: A randomised controlled trial. *British Medical Journal*, *312*, 22–26.
- Taylor, R. R., Friedberg, F., & Jason, L. A. (2001). *A clinician's guide to controversial illnesses: Chronic fatigue syndrome, fibromyalgia, and multiple chemical sensitivities*. Sarasota, FL: Professional Resource Press.
- Taylor, R. R., Jason, L. A., & Schoeny, M. E. (2001). Evaluating latent variable models of functional somatic distress in a community-based sample. *Journal of Mental Health*, *10*, 335–349.
- Van Der Linden, G., Chalder, T., Hickie, I., Koschera, A., Sham, P., & Wessely, S. (1999). Fatigue and psychiatric disorder: Different or the same? *Psychological Medicine*, *29*, 863–868.
- Vercoulen, J. H. M. M., Swanink, C. M. A., Galama, J. M. D., Fennis, J. F. M., Jongen, P. H. J., Hommes, O. R., Van Der Meer, J. W. M., & Bleijenberg, G. (1998). The persistence of fatigue in chronic fatigue syndrome and multiple sclerosis: Development of a model. *Journal of Psychosomatic Research*, *45*, 507–517.
- Ware, J. E., & Sherbourne, C. D. (1992). The MOS 36-item short-form health survey (SF-36): Conceptual framework and item selection. *Medical Care*, *June*, 473–483.

- Ware, J. E., Snow, K. K., Kosinski, M., & Gandek B. (2000). *SF-36 Health Survey: Manual and interpretation guide*. Lincoln, RI: Quality Metric Incorporated.
- Werf, van der, S. Prins, J., Servaes, P., Meer, van der, J. & Bleijenberg, G. (2003). The relation between daily fatigue patterns and physical activity in chronic fatigue syndrome. Manuscript submitted for publication.
- Werf, van der, S. Prins, J., Vercoulen, J., Meer, van der, J. & Bleijenberg, G. (2000). Identifying physical activity patterns in chronic fatigue syndrome using actigraphic assessment. *Journal of Psychosomatic Research*, 49, 373–379.
- Wessely, S., Hotopf, M., & Sharpe, M. (1998). *Chronic fatigue and its syndromes*. New York, NY: Oxford University Press.
- Wessely, S., & Powell, R. (1989). Fatigue syndromes: A comparison of chronic 'postviral' fatigue with neuromuscular and affective disorders. *Journal of Neurology, Neurosurgery and Psychiatry*, 52, 940–948.