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Premorbid "overactive" lifestyle in chronic fatigue syndrome and fibromyalgia An etiological factor or proof of good citizenship?

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Abstract

Objective: In a former study, we have shown that patients suffering from chronic fatigue syndrome (CFS) or chronic pain, when questioned about their premorbid lifestyle, reported a high level of "action-proneness" as compared to control groups. The aim of the present study was to control for the patients' possible idealisation of their previous attitude towards action. **Methods:** A validated Dutch self-report questionnaire measuring "action-proneness" (the HAB) was completed by 62 randomly selected tertiary care CFS and fibromyalgia (FM) patients, as well as by their significant others (SOs). **Results:** HAB scores of the

patients and those of the SOs were very similar and significantly higher than the norm values. Whether or not the SO showed sympathy for the patient's illness did not influence the results to a great extent. SOs with a negative attitude towards the illness even characterized the patients as more "action-prone." **Conclusions:** These results provide further support for the hypothesis that a high level of "action-proneness" may play a predisposing, initiating and/or perpetuating role in CFS and FM. © 2001 Elsevier Science Inc. All rights reserved.

Keywords: Action-proneness; Chronic fatigue syndrome; Fibromyalgia; Overactive lifestyle; Stress

Introduction

Two overlapping syndromes, chronic fatigue syndrome (CFS) and fibromyalgia (FM), remain both enigmatic and controversial despite considerable research efforts [1,2]. Many authors believe that they represent the same condition, although their pathophysiological mechanisms may differ [3]. With regard to the etiology, the available evidence suggests that both syndromes may be at the end of a multicausal process in which predisposing, initiating and perpetuating factors can be distinguished [4].

One of the possible psychosocial factors that may play an etiological role in both conditions is the lifestyle premorbidly adopted by the patients. Since the introduction of the "neurasthenia" concept — generally considered a "precursor" of CFS — chronic fatigue has often been linked with "overwork": CFS sufferers have been described as "overactive," "high achievers," "type-A-like," "perfectionist" or "unable to set limits to the demands of others" [5,6].

According to some investigators, chronic pain patients also frequently report a history of "overactivity," the socalled ergomania [7], but systematic research on this lifestyle characteristic remains inconclusive [8]. In CFS patients, some empirical evidence has been found for high selfreported "action-proneness" and "negative perfectionism" [9–12]. However, doubts have been formulated about the validity — and, thus, the etiological role — of high "actionproneness" in CFS because the patients' self-descriptions might be biased by retrospective idealisation [4].

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The aim of this study was to check the validity of high "action-proneness" reported by the patients by involving their significant others (SOs). In order to minimize a possible bias associated with the SO's attitude, it was also assessed whether the SO had a positive or negative attitude towards the patient's illness.

The research questions were, more concretely: (1) Is the "action-proneness" of CFS and FM patients, as judged by their SO, higher than the "action-proneness" of a norm group or other patient groups? (2) How large is the association and the agreement between the patients' self-report and the judgment by the SOs? (3) Is the association or agreement influenced by the SO's attitude towards the patient's illness?

Method

Sample and data collection

Participants of the study were recruited from a multidisciplinary screening of patients with symptoms of chronic fatigue and/or widespread musculoskeletal pain consulting at the departments of General Internal Medicine and Rheumatology at the University Hospital Leuven (Belgium). When the symptoms could not be explained by a thorough medical investigation, patients were systematically referred for extensive psychometric testing (including the HAB questionnaire; see below), a semistructured psychiatric indepth interview and effort capacity measurement.

One hundred and ninety-two consecutive attenders who met the criteria for CFS (n = 124) [13] or FM (n = 68) [14] were evaluated. From this group, a simple randomized sample of hundred patients was taken and an adapted version of the HAB questionnaire was sent to their partner or a parent when the patient was single. The questionnaire and a prepaid envelope were sent several weeks after the last consultation in order to avoid any interference with the diagnostic process.

Assessment

All patients completed the HAB, a Dutch self-report questionnaire developed by Dirken [15], containing 50 items to be qualified as "correct" or "incorrect." Typical items are: "I have always been an active and busy person," "I do not like to postpone things," "I love making a supreme effort," etc. The instrument has proven to be sufficiently reliable and valid. Applied to a Dutch norm group of 316 industrial workers, the mean HAB value is 29.4 (S.D. = 6.5). As in our earlier study [9], we reformulated all questions in the past tense to enable its use for the patients' retrospective evaluation of their "action-proneness," i.e., viewed from the premorbid situation.¹

All SOs completed an adapted HAB in which the pronoun "I" was replaced by "he" or "she," according

to the patients' gender. The SO's attitude towards the illness was systematically questioned during the semistructured psychiatric in-depth interview and qualified by the interviewer as "positive" or "negative."

Seventy-one SOs returned the questionnaire. Three of them could not be identified, whereas six were incomplete, which meant that 62 questionnaires could be interpreted. Thirty-four belonged to the CFS and 28 to the FM subgroup. There were no major differences in return pattern between these two subgroups. In addition, the positive or negative attitude towards the illness was not significantly different between the CFS and FM subgroups ($\chi^2 = 1.6935$, df = 1, P = .1931).

Statistical analysis

With respect to the first research question, we performed Student's t tests for independent groups. The second research question was handled by calculating Pearson's correlation and Cohen's weighted kappa coefficients and by using t and exact permutation tests. An analysis of variance with one "within factors" (self-report vs. other) and one "between factors" (with vs. without positive attitude) was carried out to answer the third research question. The robustness of the statistical results was checked by performing related nonparametric analyses.² Chi-square tests were used for more detailed analyses at the item level.

Analyses were conducted using SAS software [16] and StatXact [17]. Results with P values smaller than .05 were considered statistically significant. All reported P values are two-tailed.

Results

Descriptive statistics of age, sex, duration of symptoms and occupational level of the patient sample (N=62) are shown in Table 1. The mean HAB score for the sample is 35.0 (S.D.=6.83), which is significantly higher than the mean value of 29.4 (S.D.=6.5) for the norm group, t(376)=6.15, P<.0001.

The mean HAB score for the 62 SOs amounts to 34.5 (S.D. = 7.98), which is also significantly higher than the mean value for the norm group, [t(376) = 5.43, P < .0001]. Furthermore, it significantly exceeds the mean HAB score of 30.6 (S.D. = 7.70) we found in a previous study with 65 patients suffering from neurotic–dysthymic and chronic organic disorders, [t(125) = 2.78, P = .0062 [11]].

Regarding the second research question, we find a statistically significant positive linear relationship between the 62

¹ When the term "action-proneness" is used in this article, it always means "action-proneness" viewed from the premorbid situation.

² Since the nonparametric analyses confirmed the parametric analyses, only the latter are reported in this article. More detailed results can be obtained from the authors.

Table 1 Descriptive statistics on age, gender, duration of symptoms, occupational level and HAB scores

	CFS/FM ($N=62$)
Age (mean±S.D.)	37.4 ± 9.16
Gender (male/female)	17/45 (27%/73%)
Duration of symptoms (mean \pm S.D.)	33.8 ± 49.9 months
Occupation	
Blue collar	38 (61%)
White collar	24 (39%)
HAB (mean ± S.D.)	35.0 ± 6.83

HAB scores of the SOs and the corresponding patients, [Pearson's r=.47, t(60)=4.13, P<.0001]. There is also considerable agreement in level of "action-proneness": The mean HAB score for the 62 corresponding patients equals 35.0 (S.D. = 6.83), which is very similar to the value obtained for the SO paired samples, [t(61)=0.43, P=.6685]. Table 2 shows that there is 64.52% agreement (see percentages in bold) between the HAB scores of the SOs and those of the corresponding patients for the three major categories (low, medium, high) of HAB scores, and that there is only one large disagreement (high self-report, low SO score). Cohen's weighted kappa agreement coefficient for this table is .49, exact permutation P<.0001.

An analysis of variance with respondent as "within factors" (self vs. other) and attitude as "between factors" (with vs. without positive attitude) shows no main effect of respondent, [F(1,60)=1.18, P=.2819], and no main effect of attitude, [F(1,60)=1.84, P=.1796], but a statistically significant interaction effect, [F(1,60)=4.07, P=.0482].

Contrary to our expectations, SOs with a positive attitude towards the patient's illness provide lower HAB scores than the related patients, while SOs with a negative attitude towards the patient's illness provide higher HAB scores than the related patients (see Table 3).

As shown in Table 4, the correlation and agreement coefficients are affected in the same direction. There is much more association and agreement between the patients and SOs with a negative attitude towards the patient's illness, than between the patients and SOs with a positive attitude. Cohen's weighted kappa agreement coefficient for SOs with a negative attitude is .60, exact permutation P=.0012. For SOs with a positive attitude, it is only .32, exact permutation P=.0943.

Table 2 Agreement between patients' self-reported HAB scores and HAB scores for these patients by significant others

	Significant			
Self-report	Low	Medium	High	Total
Low (HAB \leq 24)	2 (3.23%)	1 (1.61%)	0 (0.00%)	3 (4.84%)
Medium (25-34)	3 (4.84%)	11 (17.74%)	11 (17.74%)	25 (40.32%)
High (HAB \geq 35)	1 (1.61%)	6 (9.68%)	27 (43.55%)	34 (54.84%)
Total	6 (9.68%)	18 (29.03%)	38 (61.29%)	62 (100%)

Table 3

Means and standard deviations for patients' self-reported HAB scores and HAB scores by significant others, separately for significant others with a positive or with a negative attitude towards the patient's illness

	Respondent					
	Patient		Others		Difference	
Attitude	М	S.D.	М	S.D.	М	S.D.
Positive $(n=30)$	34.8	5.66	32.4	7.95	2.4	7.87
Negative $(n=32)$	35.1	7.86	36.5	7.59	-1.4	7.12
Total $(N=62)$	35.0	6.83	34.5	7.98	0.5	7.98

Finally, a more detailed item analysis provides frequency tables of the different items and significant associations of items with the positive or negative attitude of the SOs.

First, irrespective of their attitude, more than 90% of the SOs agree with the following items: "he/she did not like to postpone things" and "he/she has always been a very busy person." More than 85% agree with the items: "in fact, he/ she was occupied with something all day," "mostly he/she needed a short training period," "he/she never was bored" and "if he/she would not have known how to set about his/ her work, he/she never would have been that successful." In addition, more than 85% disagree with the item: "if it would have been up for him/her, he/she would have slept far into the day."

Second, as compared to the patients, SOs with a positive attitude agree significantly less with "it happened that he/she moved too fast when he/she started a task" ($\chi^2 = 5.7109$, df = 1, P = .0169) and "at night, he/she regularly had a feeling of satisfaction that he/she had worked hard" ($\chi^2 = 4.3200$; df = 1, P = .0377).

Third, as compared to the patients, SOs with a negative attitude agree significantly less with "he/she found that working is OK, but you must forget it completely at night" ($\chi^2 = 6.3492$, df = 1, P = .0117).

Discussion

The patients' self-descriptions confirmed

In this study, we tried to validate our clinical experience and previous psychometric findings about the possible etiological role of high "action-proneness" in CFS and

Table 4

Pearson correlation and Cohen's weighted kappa coefficient for patients' self-reported HAB scores and HAB scores by significant others, separately for significant others with a positive or with a negative attitude towards the patient's illness

	Pearson's r			Cohen's κ		
Attitude	r	t	df	P value	к	P value
Positive $(n=30)$.37	2.11	28	.0437	.32	.0943
Negative $(n=32)$.58	3.87	30	.0006	.60	.0012
Total $(n=62)$.47	4.13	60	<.0001	.49	<.0001

FM. More concretely, we challenged the alternative hypothesis that patients suffering from a nonobjectifiable illness like CFS or FM might idealise their premorbid lifestyle. By describing themselves as "very action-oriented" patients could indeed prove their "good citizenship" and consequently diminish the risk of being stigmatised as a "psychiatric patient."

Our results, however, show that the HAB scores of this patient sample were not only as high as in our former study, but were also very similar to those of the patients' SOs. This conclusion can be made by comparing the total HAB scores, as well as via the item analysis. Indeed, 80–90% of the SOs agree with statements that focus on activity, drive, achievement, impatience and "to know how to set about work." This quasiunanimous agreement consistently strengthens the image of a premorbidly passionate, strong-willed, energetic and driven, i.e., high "action-prone" individual.

Moreover, this image is supported independently of the SO's attitude towards the illness. There is even a tendency for SOs showing a negative attitude to appraise the patients "action-proneness" as higher than for those who are supporting and solicitous, which rules out the possibility that the SO's personal bias would have a great influence on the results.

Methodological considerations

It should be mentioned that we investigated a tertiary care CFS/FM patient sample, implying a selection bias that requires some caution in generalising the findings. Furthermore, the relationship between the concept of "action-proneness" — as measured by the HAB — and various behavioral or personality features promoting "overactivity," such as high achievement motivation, obsessive-compulsive traits, perfectionism, type-A (like) behavior, "workaholism," self-sacrificing tendencies, alexithymia, etc. is not clear [6].

Within these limitations, however, our results do not support the hypothesis that CFS/FM patients retrospectively idealise their premorbid lifestyle or attitude towards activity. In the following paragraphs, we discuss the etiological and therapeutic relevance of these findings.

High "action-proneness" as an etiological factor

Although the etiology of CFS and FM remains controversial, most researchers considered it a complex, multifactorial process in which various forms of physical and/or psychosocial stress may play a key role [18–20]. Our present and previous studies suggest that high "actionproneness," by promoting an "overactive" lifestyle, may be one of the factors that makes people more vulnerable to CFS/FM, and also contributes to the onset and perpetuation of the illness.

High "action-proneness" as a predisposing factor

People who engage in an "overactive" lifestyle may run a greater risk of acute or chronic physical overburdening by a negligent attitude towards the body, musculoskeletal overuse or sleep deprivation. Particularly those with childhood victimization experiences often show a tendency to exceed physical limits (in work or sports) as a way of coping, i.e., to maintain self-esteem, stabilise the affective equilibrium and prevent anxiety and depression [6,9,20].

High "action-proneness" as an initiating factor

Long-lasting physical or mental stress may, in susceptible individuals, eventually lead to neuroendocrine (mainly HPA axis) and immunological dysfunctioning, paving the way for various stress-related disorders [21]. For example, in overtrained athletes, overworked laborers and teachers suffering from burnout, low morning cortisol has been found [22,23], which is consistent with findings of HPA axis hyporeactivity in CFS and FM [24-26]. In the same vein, slower wound healing and a higher susceptibility for infections has been demonstrated in caregivers for Alzheimer's and schizophrenic patients, respectively [27,28], suggesting that chronic life burden, by lowering immunocompetence, may play an initiating role in CFS [29]. Moreover, it has been hypothesized that stress-induced HPA axis alterations may contribute to the neuronal sensitization of central pain mechanisms, resulting in the typical "general pain hypersensitivity" of FM [30].

High "action-proneness" as a perpetuating factor

Persons who used to strive frenetically for achievement, approval or perfection may fall prey to a "self-handicapping strategy" when facing serious functional limitations [31]. This means that cognitive mechanisms (e.g., somatic attribution of failure or unrealistic goal setting when becoming ill) [32], psychodynamic processes (e.g., reversal of "counterdependence" into exaggerated dependence) [33], as well as operant learning factors may contribute to the maintenance of illness behavior. In some patients, moreover, being deprived of "overactivity" as their favourite coping strategy, fatigue and pain may be reinforced by chronic sympathetic arousal, hyperventilation and disturbances of the sleep–wake cycle.

Therapeutic implications

Given the presumable etiological role of high "actionproneness" in CFS and FM, lifestyle adjustment should always be a central goal in cognitive-behavioral programs for these patients. This is evidently not an easy task for individuals whose self-esteem has been strongly dependent on high achievement and appreciation from others, which may still drive them to periodic "outburst of activity" when feeling somewhat better. However, patients can be helped to better recognize and respect their limits and substitute their previous "overactive" lifestyle by a more balanced activity-rest schema [34]. For those with a history of victimization or serious personality disorders, experiential, interpersonal or psychodynamic therapy may also be considered [35]. These therapeutic strategies may enable CFS and FM patients to reduce chronic stress by making more realistic and priority-based life choices, learning more adequate ways of dealing with negative life events and distressing emotions, basing their self-esteem not solely on achievement-oriented activities, working less obsessively and perfectionistically, responding more assertively to the others' demands and expectations and expressing their own needs more directly and explicitly.

Conclusion

The results of this study provide further support for the hypothesis that high "action-proneness" and an associated "overactive" lifestyle may be one of the factors that play a predisposing, initiating, as well as perpetuating role in CFS and FM. The ways leading from high "action-proneness" or "overactivity" to chronic fatigue and pain may involve psychological, as well as physiological aspects. With respect to the latter, recent research has opened exciting perspectives to better understand how severe life burden might eventually give rise to a long-lasting dysfunction of the stress system. In words that reflect the story of many CFS and FM patients: how the exhausted fighter eventually capitulates.

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