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Long-term follow-up after cognitive behaviour therapy for chronic fatigue syndrome☆

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ABSTRACT

Objective: Cognitive behaviour therapy (CBT) is an effective treatment for chronic fatigue syndrome (CFS). Main aim was to determine whether treatment effects were maintained up to 10 years after treatment.
Methods: Participants (n = 583) of previously published studies on the effects of CBT for CFS were contacted for a long-term follow-up assessment. They completed questionnaires on main outcomes fatigue severity (CIS) and physical functioning (SF-36). The course of these outcomes since post-treatment assessment was examined using mixed model analyses.
Results: Between 21 and 125 months after finishing CBT, 511 persons (response rate 88%) completed a follow-up assessment. At follow-up, mean fatigue severity was significantly increased to 37.60 (SD = 12.76) and mean physical functioning significantly decreased to 73.16 (SD = 23.56) compared to post-treatment assessment. At follow-up still 37% of the participants had fatigue scores in the normal range and 70% were not impaired in physical functioning.
Conclusion: Positive effects of CBT for CFS on fatigue and physical functioning were partly sustained at long-term follow-up. However, a subgroup of patients once again reported severe fatigue, and compromised physical functioning. Further research should elucidate the reasons for this deterioration to facilitate the development of treatment strategies for relapse prevention.

1. Introduction

Patients with chronic fatigue syndrome (CFS) suffer from medically unexplained, severe fatigue leading to substantial disability [5]. According to the US Centers for Disease Control and Prevention (CDC), persons with CFS have experienced fatigue for at least six months, and their fatigue must be accompanied by other symptoms [5,6]. Cognitive behaviour therapy (CBT) is an effective treatment for CFS and has been developed based on a model of perpetuating factors [7]. This model assumes that behaviour- and fatigue-related beliefs maintain fatigue and disability. CBT aimed at these cognitive-behavioural factors significantly reduces fatigue and disability [8,9] and a minority of patients are fully recovered post-treatment [3,10]. However, little is known about the long-term effects of CBT for CFS [8]. Research into the long-term efficacy of CBT for other disorders has shown that sustainment of treatment effects is not self-evident [11,12].

For CFS, several studies that investigated short-term treatment effects found sustained effects up to eight months after the end of treatment [13–15]. Two studies had a longer follow-up period [16,17]. In the smaller study of Deale et al. [16], most patients reported sustained improvement at five-year follow-up. However, significantly more patients were severely fatigued and fewer patients reported good physical functioning at long-term follow-up compared to short-term follow-up. More recently, Sharpe et al. [17] found sustained positive effects of CBT on fatigue and physical functioning at a median follow-up period of 19 months.

☆ This study was conducted at the Radboud university medical center, Nijmegen, the Netherlands.

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Previous research showed that CFS patients with somatic co-morbidity [3] and more pain [18] have less favourable outcomes following CBT. Mental health problems are more prevalent in CFS patients and are known to be associated with fatigue [19]. All of these factors might not only influence treatment outcome but especially when they occur after end of treatment also influence long-term effects of CBT.

1.1. Aims of the study

In this study, we examined whether the positive effects of CBT on fatigue severity and physical functioning were maintained up to 10 years after the end of treatment. We also included participants' short-term follow-up data in the analyses. In order to determine what factors might be influencing treatment outcomes, we also examined whether somatic co-morbidity that occurred since the end of treatment and was still present, participants' pain and mental health at the time of the long-term follow-up, and several other patient and treatment characteristics were associated with the course of fatigue and physical functioning over time.

2. Methods

2.1. Study design and participants

Participants from four published studies that had tested the effects of CBT were contacted for a long-term follow-up assessment. These studies had been conducted at the Radboud university medical center in The Netherlands (see [1–4]). All patients were consecutively referred and met CDC criteria for CFS when included in the original study [5,6]. Patients were both severely fatigued and severely impaired, operationalized as scoring ≥ 35 on the Fatigue Severity subscale of the Checklist Individual Strength (CIS) [20] and a weighted score of ≥ 700 on the Sickness Impact Profile (SIP) [21], respectively. All patients had signed a written informed consent. Participants had received CBT in different formats: individual therapy, group therapy or stepped care [1–4,22–24]. The wait-list group of the group CBT study was not assessed at long-term follow-up because the effect of CBT following the waiting list was not published {Wiborg, 2015 #1624}. We only included patients who had received CBT in the published studies.

The stepped care consisted of a minimal intervention based on the CBT protocol (booklet with instruction combined with email contact with a therapist), followed by individual CBT if the patient had not profited enough from the minimal intervention [4] (see Fig. 1). Two studies were RCT's [2,4] and in both the principle of intention-to-treat was applied. The two other studies were cohort studies [1,3] which enrolled consecutively referred patients who started treatment.

All CBT formats are aimed at changing fatigue perpetuating beliefs and behaviour [23]. CBT starts with informing patients about the cognitive behavioural model of CFS. The patient formulates treatment goals aimed at recovery, defined as no longer being severely fatigued and disabled. Patients learn to regulate their sleep-wake pattern, to shift attention away from fatigue and to formulate helpful beliefs with respect to fatigue and the ability to become more active. All patients follow a graded activity program. Patients who are relatively active, characterised by a variable level of physical activity, first learn to divide their activities more evenly before they start increasing their level of activity, usually by walking or cycling. Low active patients, who are characterised by a very low level of physical activity, immediately start increasing their physical activity. If patients believe that they are able to increase activity, they start realising their goals, including resumption of work. Therapy ends with an evaluation.

Patients were assessed at baseline, post-treatment and at short-term follow-up about six months after finishing their treatment. The effects of CBT at post-treatment have been published. Previous studies have shown that at short-term follow-up the effects of CBT were sustained. The present study focused on the results of the long-term follow-up, but we also examined data on short-term follow-up in order to compare the data with previous short-term follow-up studies. Short-term follow-up assessments were previously collected as part of the routine clinical care. Most short-term follow-up studies on CBT for CFS conducted a follow-up assessment between six and eight months after treatment [13–15] and a large follow-up study after a median of 19 months after the final outcome assessment [17]. We defined short-term follow-up as an assessment between three and eighteen months after post-treatment assessment. Long-term follow-up was defined as an assessment that took place more than eighteen months after post-treatment assessment.

2.2. Procedures

A research assistant contacted potential participants by telephone, and once they agreed to fill in a long-term follow-up assessment, they received an invitation letter and questionnaires by mail or e-mail (with a link to the complete questionnaires online). The questionnaire was sent again if the participant did not return it by post or complete it online within two weeks. If only a postal or e-mail address was known, the invitation letter was sent without an initial phone call. If no address was known, the municipal registration was consulted. When participants refused to fill in questionnaires, they were asked to complete the subscale fatigue severity of the CIS by phone. Before participating in one of the previous studies, all the participants had already provided written informed consent. These studies and the follow-up study were approved by the local medical ethical committee [1–4].

2.3. Measures

The long-term follow-up assessment consisted of three parts and took a maximum of 15 min to complete. A *general questionnaire* contained items on the participant's work situation, somatic co-morbidity, life events, CFS and treatment for fatigue.

Fatigue was assessed with the Fatigue Severity subscale of the CIS [20] consisting of eight items scored from one to seven. The subscale score varies between eight and 56. A score below 35 indicates fatigue in the normal range, a score of 35 or higher indicates severe fatigue [25]. The CIS is a reliable and valid instrument for CFS [26].

Physical functioning, mental health and bodily pain were assessed with the respective subscales of the SF-36 [27]. Weighted subscale scores range from 0 to 100, higher scores indicate a better health status. A score of ≥ 65 was used as a criterion for physical functioning in the normal range [2]. The SF-36 is a reliable and valid instrument for different patient populations [28].

2.4. Statistical analyses

All analyses were performed with IBM SPSS Statistics 20. The threshold for significance was $p < 0.05$ (two-tailed). Sample characteristics were described using numbers, percentages and mean scores. With the use of independent *t*-tests, non-participants and participants were compared with regard to age and scores on the subscale fatigue severity and physical functioning at post-treatment and short-term follow-up.

Each participant had fatigue and physical functioning data of maximal four measurement points: baseline, post-treatment, short-term follow-up and long-term follow-up. Mean scores on all points were displayed in a figure. The long term follow-up was subdivided into different time clusters of the number of months between post-treatment and follow-up. Mean fatigue and physical functioning was displayed for the time clusters in a separate figure.

To examine the course of fatigue and physical functioning scores over time, linear mixed model analyses were applied, where the repeated measures at post-treatment, short and long-term follow-up were clustered within the subject. Mixed models are by default able to