A self-management programme for individuals with chronic fatigue syndrome:

Development, piloting and evaluation

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Papers
1. Preface

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List of papers


III. Pinxsterhuis, I, Strand EB, Sveen U. Coping with chronic fatigue syndrome: A review and synthesis of qualitative studies. (submitted 8th of December 2014)

Abstract

Background

Chronic fatigue syndrome (CFS) is a disabling condition that affects different aspects of everyday life, including social and vocational participation. There is currently no known cause, cure or widely accepted treatment for CFS. Although some patients recover, CFS remains a chronic condition for the majority of affected patients, who thus must learn how to cope with the illness. Recent research has revealed that receiving help to cope with CFS during the early phases of the illness prevents relapses and results in a better prognosis. However, several studies have shown that patients receive insufficient information, support and help from their general practitioners (GP’s) and other healthcare professionals after diagnosis.

In Norway, the structure of the healthcare system is guided by the Coordination Reform, which promotes access to good and equal healthcare services regardless of an individual’s place of residence. The reform emphasises the need to establish patient education programmes in primary healthcare. However, several studies have indicated that patient education alone is not sufficient to improve coping and should be complemented by self-management education. None of the known patient education and self-management programmes for individuals with CFS can be implemented in Norway’s primary healthcare system without modifications because of the specific organisation of the system and the limited availability of primary care professionals with competence in CFS.

Aim

The overall aim of this study was to develop and evaluate a group-based self-management programme for adults with CFS that promotes coping with the illness and that can be implemented in a primary healthcare setting in Norway.

Material and methods

We were inspired by the framework developed by the Medical Research Council in the United Kingdom when developing, piloting and evaluating the self-management programme in this study.
Study I was a qualitative study that aimed to elicit the participants’ experiences with the patient education programme for individuals with CFS at Oslo University Hospital and their views regarding the usefulness of the programme. Focus groups were conducted immediately and nine months after participation in the programme. All of the patients that participated in the CFS patient education programme from February to April 2010 were invited to participate in this study. The final sample comprised eight women and two men with CFS who ranged in age from 32-57 years. The interviews were analysed using thematic analysis.

Study II was a qualitative study that aimed to develop a group-based self-management programme for individuals with CFS by using the participants’ experiences with the initial version of the programme. This initial programme aimed to promote coping with CFS in a primary healthcare setting in Norway. Focus groups were conducted immediately and four months after participation in the programme. The participants were recruited from a mid-sized town in southeast Norway and its surrounding municipalities in January and February 2011. All of the participants in the self-management programme were invited to participate in the study. The final sample comprised five women and one man with CFS who ranged in age from 28-46 years. The interviews were analysed using thematic analysis.

Study III was a review and synthesis of qualitative studies that focused on coping with CFS. The aim of this study was to identify factors that promote coping with CFS and to provide recommendations for adequate interventions in this patient group. Article selection was initiated by searching major scientific databases for relevant studies published before October 2013. Fifteen studies were included in the review. The studies were analysed using meta-ethnography.

The aim of study IV was to evaluate the effectiveness of the developed self-management programme in a randomised controlled trial. Participants were recruited from four mid-sized towns in southern Norway, two suburbs of Oslo and their surrounding municipalities from November 2011 to January 2012. A total of 146 participants with CFS were randomly assigned to the intervention or control group. Random assignment was done using a computerised randomization list that was created in blocks of six with the municipality as the stratifying variable. A total of 137 participants remained in the study at baseline. Assessments of physical functioning, fatigue severity, self-efficacy, health status and acceptance were performed at baseline and at six-month and one-year post-intervention. The paired-samples t-
test was used for within-group comparisons and a mixed-model analysis for repeated measures was performed to investigate the effects of the intervention compared with controls.

**Main results**

Initially, the participants in the patient education programme at Oslo University Hospital (paper I) experienced confusion and insecurity about coping with the illness. Participation in the programme appeared to promote understanding, acceptance and coping through gaining greater knowledge, learning coping strategies, and exchanging experiences, as well as receiving understanding and acceptance. The results of this study were taken into account for the development of the initial version of the self-management programme.

The initial version of the self-management programme developed in study II appeared to be a feasible intervention that could be provided in a primary healthcare setting, although several modifications regarding the content and practical organisation of the programme were proposed. The experiences of the group leaders and participants were taken into account for the development of the final programme.

The findings of the review (paper III) suggested that coping strategies, including activity management and the use of cognitive and emotional strategies, and psychological processes, such as acceptance and the rebuilding of identities and lives, may promote coping with the illness. The use of adequate coping strategies appeared to be facilitated by progress in psychological processes. Coping with CFS appeared to be facilitated mainly by self-management and complemented occasionally by treatment and social support. The findings indicated that interventions that focus on coping with CFS may include the use of adequate coping strategies and may take into account progress in psychological processes. The results of this study were used in the development of the final version of the self-management programme.

In study IV, no significant differences were identified within or between the groups in physical functioning. At the six-month follow-up, a significant difference was found between the two groups concerning fatigue severity in favour of the control group (p = 0.039) and concerning self-efficacy in favour of the intervention group (p = 0.039). These significant differences were not sustained at the one-year follow-up. Acceptance improved significantly in the intervention group at the six-month (mean change = 0.9, p = 0.006) and one-year (mean
change = 0.7, p = 0.030) follow-ups and in the control group at the six-month follow-up (mean change = 1.1, p = 0.001). The dropout rate was low (13.9 %) and the median number of attended sessions was high (seven of maximum eight).

**Conclusions**

The programme that was evaluated in study I was found to be a beneficial intervention. However, the participants expressed a need for guidance or follow-up after participation in the programme to maintain their use of strategies that help them cope with their illness.

Consistent with the experiences of the participants in study II and the results of study III, we developed the final self-management programme which was provided in eight biweekly sessions and led by juxtaposed peer counsellors and occupational therapists.

The evaluation of the developed self-management programme for people with CFS (study IV) suggested that the programme had a stabilising effect on fatigue severity and on self-efficacy at the six-month follow-up compared with people with CFS receiving usual care. The low dropout rate and the high number of sessions attended may indicate that this programme is an acceptable and helpful intervention. Longer-term quantitative and qualitative follow-up studies are needed before firm conclusions can be drawn regarding the effectiveness of our self-management programme.

There are challenges inherent in designing studies to reveal effects of complex interventions. Therefore, a process evaluation and longer-term follow-up studies are needed before we can draw firm conclusions about the effectiveness of our programme. In addition, research is needed to evaluate the cost-effectiveness of the programme. The poor development of measures of complexity in healthcare continues to present a challenge.
Sammendrag

Bakgrunn

Kronisk utmattelsessyndrom (CFS) er en invalidiserende tilstand som påvirker forskjellige aspekter av daglig livet, inkludert sosialt samvær og deltakelse i arbeidslivet. Per dags dato finnes det ingen kjent årsak, behandling eller allmenn akseptert behandling for CFS. Til tross for at noen pasienter blir helt friske, forblir sykdommen for de fleste en kronisk tilstand som de må lære seg å leve med. Forskning har vist at å få hjelp til å mestre hverdagen med CFS i tidlig fase av sykdommen kan forebygge tilbakefall og føre til en bedre prognose. Flere studier har likevel antydet at pasienter mottar utilstrekkelig informasjon, støtte og hjelp fra fastleger og annet helsepersonell etter å ha fått CFS-diagnosen.

Samhandlingsreformen tar utgangspunkt i tilgang til gode og likeverdige helse- og omsorgstjenester, uavhengig av bosted. Den legger vekt på betydningen av mestring av eget liv og nevner etablering av lokale lærings- og mestringstilbud som et av flere mulige tiltak. Flere studier har vist at pasientundervisning alene ikke er nok for å oppnå bedre mestring og at pasientundervisning bør bli kombinert med selvhjelpsteknikker. Ingen av de kjente mestringskursetene for personer med CFS kan bli implementert i norsk helsevesen uten tilpasninger på grunn av den spesifikke organiseringen av norsk helsevesen og begrenset tilgang til helsepersonell i primærhelsetjenesten med kompetanse på CFS.

Mål

Overordnet mål for studien var å utvikle og evaluere et selvhjelpskurs for voksne med CFS som fører til bedre mestring av sykdommen og som kan bli implementert i primærhelsetjenesten.

Materiale og metode

I denne studien ble utvikling, utprøving og evaluering av selvhjelpskurset inspirert av et rammeverk som er utviklet av Medical Research Council i Storbritannia.

Studie 1 var en kvalitativ studie som hadde som mål å belyse deltakernes erfaringer med mestringskurset for personer med CFS ved Oslo universitetssykehus og deres syn på nytteverdien av kurset. Fokusgruppeintervjuer ble gjennomført direkte etter og ni måneder etter deltakelse på kurset. Alle deltakerne som deltok på mestringskurset i perioden februar til
April 2010 ble invitert til å delta i studien. Utvalget som deltok i fokusgruppeintervjuene bestod av åtte kvinner og to menn med CFS i alderen 32-57 år. Intervjuene ble analysert ved hjelp av tematisk analyse.


Resultater

Deltakerne på mestringskurset på Oslo Universitetssykehuset (artikkel I) erfarte før deltakelse på kurset forvirring og usikkerhet i forhold til mestring av sykdommen. Deltakelse på kurset viste seg å kunne fremme forståelse, akseptering og mestring gjennom å tilegne seg mer kunnskap, lære mestringsstrategier og erfaringsutveksling, samt å møte forståelse og aksept. Resultatene av denne studien ble brukt i utviklingen av den første versjonen av selvhjelpskurset.

Den første versjonen av selvhjelpskurset som ble utviklet i studie II viste seg å være en gjennomførbar intervensjon som kunne bli organisert i primærhelsetjeneste, til tross for at flere endringer ble foreslått vedrørende innhold og den praktiske organiseringen av kurset. Kursledernes og deltakernes erfaringer ble brukt i utviklingen av den endelige versjonen av kurset.

Resultatene av kunnskapsoppsummeringen (artikkel III) antydet at mestringsstrategier, som aktivitetsregulering og bruk av kognitive og emosjonelle strategier, i tillegg til psykologiske prosesser, som akseptering og gjenoppbygging av identiteter og liv, kan fremme mestring av sykdommen. Bruk av adekvate mestringsstrategier viste seg å bli lettere når pasienter opplevde framgang i psykologiske prosesser. De inkluderte studiene antydde at mestring av CFS særlig ble fremmet gjennom selvhjelp, iblant i kombinasjon med behandling og sosial støtte. Funnene indikerte at intervensjoner som fokuserer på mestring av CFS bør inkludere bruk av adekvate mestringsstrategier og bør ta hensyn til utviklinger i psykologiske prosesser. Resultatene av denne studien ble brukt i utviklingen av den endelige versjonen av selvhjelpskurset.

Ingen signifikante forskjeller ble funnet i den randomiserte kontrollerte studien (studie IV) mellom og innenfor gruppene når det gjelder fysisk funksjon. Ved seks-måneders oppfølgelse ble en signifikant forskjell funnet mellom de to gruppentens vedrørende utmatting i favor av kontroll gruppa (p = 0,039) og vedrørende self-efficacy i favor av intervensjonsgruppa (p = 0,039). Disse signifikante forskjellige ble ikke opprettholdt etter ett år. Intervensjonsgruppa viste signifikant forbedring vedrørende akseptering etter seks måneder (gjennomsnitts forskjell = 0,9, p = 0,006) og ett år (gjennomsnitts forskjell = 0,7, p = 0,030) og i kontrollgruppa etter seks måneder (gjennomsnitts forskjell = 1,1, p = 0,001). Studien hadde lavt frafall (13,9 %) og høy oppslutning (median = syv av totalt åtte kursdager).
**Konklusjoner**

Mestringskurset som ble evaluert i studie I ble opplevd å være en nyttig intervensjon. Deltakerne gav likevel uttrykk for at de trengte veiledning eller oppfølging etter deltakelse på kurset for å kunne opprettholde deres bruk av mestregingsstrategier.

I tråd med erfaringene fra deltakerne i studie II og resultatene av studie III utviklet vi den endelige versjonen av selvhjelpskurset som bestod av åtte samlinger som ble organisert annenhver uke. Kurset ble ledet av likestilte brukerrepresentanter og ergoterapeuter.

Evaluering av det utviklede selvhjelpskurset for personer med CFS (studie IV) antyder at kurset hadde en stabiliserende effekt på utmattelse og self-efficacy etter seks måneder sammenlignet med personer med CFS som fikk vanlig oppfølgning. Lavt frafall og høy oppslutning kan tyde på at programmet er en akseptabel og nyttig intervensjon. Langvarige kvantitative og kvalitative oppfølgingsstudier trenges før vi kan trekke endelige konklusjoner vedrørende effektiviteten av vårt selvhjelpskurs.

Det er utfordrende å designe studier for å måle effekter av komplekse intervensjoner. Derfor er det behov for en prosessevaluerings og langvarige oppfølgingsstudier før vi kan trekke endelige konklusjoner vedrørende effektiviteten av vårt selvhjelpskurs. I tillegg trenges det forskning for å evaluere kostnadseffektiviteten av kurset. Det er en utfordring at det er mangel på gode instrumenter som kan måle kompleksiteten i intervensjoner.
List of abbreviations

CBT  Cognitive behaviour therapy
CDC  Centers of Disease Control and Prevention in the United States of America
CFS  Chronic fatigue syndrome
FSS  Fatigue severity scale
GET  Graded exercise therapy
GP   General Practitioner
ICQ  Illness Cognition Questionnaire
MCS  Mental component summary
ME   Myalgic encephalopati/encephalomyelitis
PCS  Physical composite summary
SF-36 Medical Outcomes Study 36-item Short Form Questionnaire
2. Introduction

Chronic fatigue syndrome (CFS), also known as myalgic encephalopathy/encephalomyelitis (ME), is a disabling condition that affects various aspects of everyday life, including social and vocational participation (1, 2). Living with the illness may lead to reduced quality of life (3), identity crisis (1, 4), and social isolation (1-4). CFS is characterised by 6 months or more of unexplained fatigue accompanied by a range of additional physical and cognitive symptoms (5).

CFS has no known cause. Because there are no specific diagnostic tests, the diagnosis is based on the description of symptoms, the patient’s functional level and the exclusion of alternative diagnoses. The prevalence of CFS is estimated to be 0.0 - 7.6 % (6), with a substantially higher prevalence among women (7). Currently, there is no known cure or widely accepted treatment for CFS. Although some patients recover, CFS remains a chronic, debilitating condition for the majority of the patients (8, 9); thus, these patients must learn how to cope with the illness.

Recent research has revealed that receiving help to cope with CFS at an early phase of the illness prevents relapses and results in a better prognosis (10). However, several studies have shown that patients receive insufficient information, support and help from their GP’s and other healthcare professionals after diagnosis (1, 11-16).

In Norway, the structure of the healthcare system is guided by the Coordination Reform, which promotes access to good and equal healthcare services regardless of an individual’s place of residence. The reform emphasises coping and the need to establish patient education programmes in primary healthcare as one of several potential actions (17). Some studies have shown that patient education programmes appear to be beneficial for people with CFS (18, 19). However, several studies have indicated that patient education alone is not sufficient to improve coping (20, 21) and should be complemented by self-management education (22). A self-management programme allows patients to identify their problems and provides techniques to help patients make decisions, take appropriate actions, and alter these actions as they encounter changes in their circumstances or during the course of their illness (22). The CFS empowerment project, which was conducted in the United States of America, included a self-management programme for patients with CFS that showed positive effects regarding symptom severity (23), quality of life (23, 24), and resource acquisition (25).

None of the known patient education and self-management programmes for
individuals with CFS can be implemented in primary healthcare in Norway without modifications because of the specific organisation of the Norwegian healthcare system and the limited availability of primary care professionals with competence in CFS. Therefore, the overall aim of this study was to develop and evaluate a group-based self-management programme for individuals with CFS that promotes coping with the illness and that can be implemented in a primary healthcare setting in Norway.
3. Aims of the study

The overall aim of this study was to develop and evaluate a group-based self-management programme for individuals with CFS that promotes coping with the illness and that can be implemented in a primary healthcare setting in Norway. The specific aims were as follows:

1. To elicit participants’ experiences with the patient education programme for individuals with CFS at Oslo University Hospital, and their views regarding the usefulness of the programme immediately and nine months following participation in the programme (paper I).

2. To develop a group-based self-management programme for individuals with chronic fatigue syndrome (CFS) by using the participants’ experiences with the initial version of the programme, which intended to promote coping with the illness in a primary healthcare setting in Norway (paper II).

3. To identify factors that promote coping with chronic fatigue syndrome (CFS) and to provide recommendations for adequate interventions in this patient group (paper III).

4. To evaluate the effectiveness of the developed self-management programme in a randomised controlled trial (paper IV).
4. Background

4.1 Defining chronic fatigue syndrome
The diagnosis of CFS is based on the description of symptoms, the patient’s functional level and the exclusion of alternative diagnoses. At least 20 different sets of diagnostic criteria for CFS have been developed since 1988 (6); the CDC-1994 case definition of CFS (5) is the most often used in research (6). According to this case definition, CFS is characterised by unexplained chronic fatigue of new onset and four or more of the following symptoms lasting at least six months: impaired short-term memory and concentration, sore throat, swollen lymph nodes, muscle pain, multi-joint pain, headaches of a new type, unrefreshing sleep, and post-exertional malaise lasting more than 24 hours. The CDC-1994 case definition was created primarily to standardise research and was considered inappropriate for making clinical diagnoses (26). Therefore, the Canadian ME/CFS case definition was developed to provide a conceptual framework for clinical diagnoses (26). Compared with the CDC-1994 case definition (5), the Canadian ME/CFS case definition requires a greater number of prominent symptoms (26). According to the Canadian ME/CFS case definition, CFS is characterised by unexplained chronic fatigue of new onset, post-exertional malaise and/or fatigue lasting more than 24 hours. In addition, a CFS diagnosis requires sleep dysfunction, pain and neurocognitive impairments, as well as autonomic, neuroendocrine and/or immune impairments. The symptoms must have persisted for at least six months (26).

Symptom severity and daily activity level may be used to classify patients into subgroups. Patients with mild CFS will present an approximately 50 % reduction in their premorbid activity level. Patients in the moderate subgroup will be mostly housebound. Patients with severe CFS will be mostly bedbound, and the very severely ill patients will be bedbound and will require help to perform daily activities (27).

4.2 Impact of chronic fatigue syndrome on occupational performance and health status
CFS is a disabling condition because of the associated severe and enduring physical and cognitive impairments. The illness may affect different aspects of everyday life, including the performance of daily activities, mobility, communication, learning abilities, careers, finances, and personal relationships (2, 28). In addition, people with CFS may experience stigmatisation (2, 29). Patients face stigma when they must address questioning about the veracity, morality, and accuracy of their symptom descriptions (29) and with the
psychologising of symptoms by doctors, family member, and friends (2). Several studies have revealed that living with the illness may lead to reduced quality of life (3), identity crisis (1, 4, 28), and social isolation (1-4).

4.3 Explanatory models of chronic fatigue syndrome
CFS has no known cause. Therefore, there are divergent views regarding the cause of the illness, which may explain the development of different case definitions. Friedberg and Jason (30) delineated four explanatory models of CFS: 1) an immune activation model that views CFS symptoms as being caused by chronic activation of the immune system; 2) a symptom avoidance model that conceptualises the illness as triggered by a transitory acute infection that is maintained by psychological factors; 3) an illness reactivity model that interprets psychological disorders as secondary reactions to a biological illness; and 4) a conversion model that views psychological factors as the primary cause of the syndrome.

4.4 Non-pharmacological interventions for chronic fatigue syndrome
Currently, there is no known cure or widely accepted treatment for CFS. Treatment strategies for the illness include psychological, physical and pharmacological interventions. The most commonly used non-pharmacological treatment strategies are cognitive behaviour therapy (CBT), graded exercise therapy (GET) and pacing.

Cognitive behaviour therapy (CBT) facilitates the identification of unhelpful, anxiety-provoking thoughts, and challenges these negative thoughts and the underlying dysfunctional assumptions through ‘hypothesis-testing’. CBT combines a graded increase in activity with a psychological approach that addresses the thoughts and beliefs about CFS that may impair recovery (31). Several randomised controlled trials have been conducted to assess the effectiveness of CBT. A Cochrane review (31) revealed that CBT is more effective than usual care for reducing the symptoms of fatigue. However, there was found a lack of evidence supporting the comparative effectiveness of CBT alone or in combination with other treatments. A comprehensive survey conducted by the ME Association in the United Kingdom (32) revealed that 25.9 % of the respondents who had tried CBT had improved, whereas 19.6 % had worsened.

Graded exercise therapy (GET) is based on theories that assume that CFS is perpetuated by reversible physiological changes caused by deconditioning and avoidance of activity. The aim of GET is to gradually return the patient to appropriate physical activity levels, reverse the deconditioning, and thereby reduce fatigue and disability (33). GET consists of an aerobic activity that uses the major large muscles of the body, such as walking,
cycling or swimming (34, 35). GET consists of establishing a baseline of achievable physical exercise, followed by a graded increase in the duration and intensity of physical activity (34). A Cochrane review (36) showed evidence that some patients may benefit from GET and no evidence that exercise therapy worsens outcomes on average. However, the previously mentioned survey (32) revealed that only 22.1% of the respondents who had tried this treatment strategy had improved, whereas 56.5% had worsened.

*Pacing* is an approach in which patients are encouraged to be as active as possible within the limits imposed by the illness (37). Pacing is based on the assumption that the symptoms reflect a pathological disturbance that is not reversible by increasing the level of activity. Symptoms are regarded as warning signs to be ‘listened to’ and patients are required to terminate an activity before or at the first sign of post-exertional malaise (37, 38). Pacing approaches may be guided by the Energy Envelope Theory, which is based on the hypothesis that avoiding over-exertion by keeping energy expenditures within the ‘envelope’ of perceived energy levels may prevent relapses and increase energy levels and activity tolerance (30). The results of several studies support the Energy Envelope Theory (39, 40). Because pacing is not considered a treatment for the illness as a whole, Goudsmit et al. recommend offering pacing as part of a multi-component management programme (37). The survey conducted by the ME Association (32) revealed that 71.2% of the respondents who had tried this treatment strategy had improved, whereas only 4.7% had worsened.

Pacing has been included in a number of multi-component programmes that have shown promising results (23, 41), also when compared to CBT, relaxation and anaerobic activity (42). Interestingly, a recent controlled trial which compared a multi-component programme that included pacing (adaptive pacing therapy) with CBT, GET and specialist medical care found that CBT and GET were associated with less fatigue and better physical functioning, while adaptive pacing therapy had little effect (33). The different results of these two trials may be explained by their use of different pacing protocols (37).

Additional research must be conducted to compare different treatment strategies and to evaluate the effectiveness of different pacing protocols. Furthermore, additional studies are required to facilitate the development of other effective non-pharmacological interventions for people with CFS.

### 4.5 Patient education and self-management education

Given the negative impact of CFS and the lack of curative treatments, effective interventions must be developed to promote patients’ ability to cope with the illness. There are several
definitions, theories and models of coping. In the present study, ‘coping’ refers to a person’s cognitive and behavioural efforts to manage specific external and/or internal demands that are appraised as stressful (43). According to Folkman and Lazarus (43), these cognitive and behavioural efforts constantly change as individuals manage particular demands that they perceived as stressful. Coping processes may be directed toward altering the situation that is causing distress (problem-focused coping) and/or regulating the emotions that result from a stressful condition (emotion-focused coping) (43).

Drachler and colleagues (11) found that patients with CFS need 1) to make sense of symptoms, 2) information about the illness, 3) to develop strategies to manage impairments and activity limitations, and 4) to develop strategies to maintain/regain social participation. The role of patient education programmes will likely become increasingly important.

4.5.1 Traditional care versus collaborative care
In traditional care, professionals are the experts, whereas patients are passive participants. Problems are identified by healthcare professionals who tell patients what to do, and the goal is compliance with the professionals’ instructions (22, 44, 45). Patient education that imparts disease-specific information and technical skills is a form of traditional care (22). In chronic illness, however, collaborative care has become increasingly important. Collaborative care is characterised by a patient-physician relationship in which patients and physicians make health care decisions together. Professionals are considered experts about diseases, and patients are considered experts about their lives. The patient sets goals, and the professional helps the patient make informed choices (22, 44, 45). Self-management education allows patients to identify their problems and provides techniques to help patients make decisions, take appropriate actions, and alter their actions when necessary (22, 44, 45).

Several studies have indicated that patient education alone is not sufficient to improve clinical outcomes (20, 21): programmes that teach self-management skills are more effective in this regard (22). However, according to Bodenheimer and colleagues (22), self-management education complements rather than replaces traditional patient education.

4.5.2 Client-centred practice
Client-centred practice has been a central concept in collaborative care (45) and in occupational therapy since 1980, when the Canadian Association of Occupational Therapists started to focus on a client-centred approach to occupational therapy. Sumsion (46, page 308) defined client-centred practice in occupational therapy as ‘... a partnership between the client and the therapist that empowers the client to engage in functional performance and fulfil his
or her occupational roles in a variety of environments. The client participates actively in negotiating goals which are given priority and are at the centre of assessment, intervention and evaluation. Throughout the process the therapist listens to and respects the clients’ values, adapts the interventions to meet the clients’ needs and enables the client to make informed decisions.’

A review revealed that key elements of client-centred practice are power sharing between the client and therapist, partnership, effective listening and communicating, respecting the clients’ ability to make choices and decisions, and maintaining hope (47). The communication of information was found to be the central focus of a client-centred approach, because information about medication, clinical status, progress, prognosis, and processes of care will enable clients to make decisions about and take responsibility for their own health (47).

Client-centred practice has been shown to improve patients’ symptoms, functional outcomes (48, 49), satisfaction with services (50), and the ability to recall their goals (48).

4.5.3 Empowerment
Empowerment is a central concept in both collaborative care and client-centred practice. Empowerment refers to transferring power from healthcare professionals to patients, meaning that healthcare professionals must provide individuals with the knowledge and resources that they need to take control of their lives (51). It includes promoting individual self-reliance and using peer networks to help with solving problems (52).

Recently, co-production or co-creation, a concept related to empowerment, has appeared in public welfare politics (51). Co-production can be defined as ‘the provision of services through regular, long-term relationships between professionalized service providers (in any sector) and service users or other members of the community, where all parties make substantial contributions’ (53, page 847). This definition indicates that co-production is characterised by partnership and power-sharing between service providers and service users (54). Co-production is an approach that recognises the resources and competences of the individual but also empowers him or her to take responsibility for finding solutions that may serve the community (51).

4.5.4 Self-efficacy
Self-efficacy is an important concept in self-management and a core concept in Albert Bandura’s social-cognitive theory, which states that ‘perceived self-efficacy refers to beliefs in one’s capabilities to organize and execute the courses of actions required to produce given
attainments’ (55, page 3). Perceived self-efficacy affects thought processes, motivation, and affective and physiological states, all of which affect behaviour. People take action when they hold efficacy beliefs and outcome expectations that make the effort seem worthwhile.

Self-efficacy beliefs begin to develop in early childhood and are constructed from four sources of information (55):

1. Enactive mastery experiences, which are the most influential source of efficacy information and provide authentic evidence of capability based on task performances.
2. Vicarious experiences, which occur through observing role models, especially those who are similar to the individual in age, sex, race, ability, and experiences.
3. Verbal persuasion by others who convey the realistic belief that the individual is capable of the expected behaviour.
4. The individual’s interpretation of his or her own physiological and affective states, which forms the basis for an individual’s judgements of his or her own capability.

Efficacy beliefs are usually quite durable but may fluctuate in strength over time. In addition, people may have a strong sense of efficacy in one activity domain and a low sense of efficacy in other realms. Therefore, measures of personal efficacy must be tailored to specific domains of functioning (55).

Two reviews articles synthesising studies of self-efficacy-enhancing interventions for people with chronic diseases concluded that enhanced self-efficacy could have a positive impact on patients’ health status, adoption and maintenance of favourable health behaviours, and adherence to prescribed regiments (56, 57). Thus, self-efficacy may serve as a predictor of health behaviour and self-management (56, 57).

4.5.5 Self-management tasks
Corbin and Strauss (58) developed a framework for managing chronic illness. Based on their own qualitative research, they delineated three sets of self-management tasks:

1. Medically managing the condition, through regimen work, crisis prevention and handling, and symptom management.
2. Maintaining, changing, and creating new meaningful behaviours concerning everyday-life, including housekeeping, occupational work, marital work, child rearing, and recreation.
3. Dealing with the emotional sequelae of having a chronic condition that alters one’s view of the future.
If we use this framework, then self-management programmes must include content that addresses all three tasks (59).

4.5.6 Adult education

Knowles, Holton, and Swanson (60) presented six core assumptions about adult learners that distinguish them from children:

1. Adults need to know how the learning will be conducted, what learning will occur, and why they need to learn something before learning it. Therefore, it is now generally accepted that adults should be engaged in the planning and facilitation of adult learning programmes.
2. The adult self-concept moves from dependency to self-direction.
3. The learner’s growing reservoir of previous experiences provides a rich resource for learning. However, previous experiences may create biases that can both inhibit and shape new learning.
4. Adults generally become ready to learn when they experience a need to cope with a life situation or perform a task.
5. Adults’ orientation to learning is life-centred. Adults generally prefer problem-solving oriented learning, rather than subject-centred learning. Moreover, they learn best when new information is presented in a real-life context. Adult education is a process of developing increased competencies that allow the adult learner to achieve his or her full potential.
6. The most potent motivators for adult learners are internal – for example, quality of life, satisfaction, and self-esteem - rather than external.

These assumptions may provide a sound foundation for planning, implementing, and evaluating teaching programs for adults (60, 61). However, the individual characteristics of the learners – including differences in cognitive functioning, personality, and prior knowledge and beliefs – as well as situational factors - such as large group learning environments and socio-cultural influences - may impact adult learning. In addition, different subject matter may require different learning strategies; for example, self-directed learning may be less suitable for learning complex technical subject matter (60). Finally, the objectives and purposes of the learning programme should be identified to provide a framework that shapes the learning experience (60).

Life-span development theories clarify and refine assumptions about adult learning by
addressing the readiness to learn aspect of the learning event. Predictable types of changes in the lives of adults often trigger a need to learn (60).

4.5.7 Individual and group education
Patient education can be provided individually or in groups. Individual teaching facilitates the sharing of confidential information and problems and is easier to adapt to the patient’s specific problems and needs than group education (44). Group education allows the participants to share knowledge and experiences with one another and helps them to process information and integrate it into their behaviours (44). Compared with individual education, group education leads to a deeper understanding and a longer retention of information, increased social support, and greater transfer of learning from one situation to another, as well as more positive interpersonal relationships, more favourable attitudes toward learning, and more active learner participation (62). Group education may also be more cost-effective than individual education (44). A review revealed that patient education may be provided individually or in groups, but it must always be based on the specific needs of the individual (63).

4.5.8 Learning activities
The Chinese proverb ‘Tell me, I forget. Show me, I remember. Involve me, I understand’ (author unknown) implies that information retention rates vary with different teaching methods (62). A lecture may be a useful method for teaching cognitive information, but lectures are most effective when they include discussion. Discussion promotes the understanding and application of knowledge and the development of certain attitudes (44). Alternative instructional methods include demonstration, role playing, return demonstration (44, 62), one-to-one instruction, gaming, simulation, role modelling, and self-instruction (62). Decisions about which methods to use may be based on a consideration of the following factors: audience characteristics, educator expertise, learning objective, and the potential for achieving learning outcomes, as well as cost effectiveness, instructional setting, and evolving technology (62). In addition, these factors may influence the use of different types of teaching tools, such as printed and audio-visual materials (44, 64).

4.5.9 Goal-setting and action planning in self-management education
Patients will be more motivated to apply themselves to learning and following health recommendations when they recognise that the given information and recommendations will help them achieve their specific goals (45). In collaborative care, the healthcare professional
guides the patient through identifying and exploring the problem, setting goals and considering the choices, choosing a plan of action, and evaluating the results (22, 44). Setting specific, achievable goals serve as motivating factor that may promote learning and health behaviour (55, 65). Patients may use short-term action plans to reach their goals. The action plan should be realistic, proposing behaviour that patients are confident they can accomplish. Using action plans makes it possible for patients to find solutions and learn problem-solving skills that may help them cope with chronic illness (22).

4.6 Previous research concerning patient education and self-management education

Holman and Lorig (66) developed a self-management programme for individuals with chronic illness based on Bandura’s self-efficacy theory. The programme included the following four characteristics of self-efficacy enhancement (55): the guided mastery of skills through weekly ‘action planning’ and feedback on progress; the modelling of self-management behaviours and problem-solving strategies; social persuasion through group support and guidance for individual self-management efforts; and the reinterpretation of physiological symptoms (67). The programme participants exhibited improved health behaviour and health status, fewer hospitalisations and shorter hospital stays at the six-month follow-up (68), and improvements in health behaviour, health status, and self-efficacy after one year (67, 69). To my knowledge no publications have addressed interventions based on self-efficacy theory for individuals with CFS.

To date, few studies have been published concerning patient education for individuals with CFS. Taylor and Jason conducted the CFS Empowerment Project, a community-based treatment programme for individuals with CFS that involved a collaboration between an academic occupational therapy department and an independent living centre in Chicago (70). This programme was designed to provide CFS patients with an opportunity to learn self-advocacy skills and to improve their quality of life, coping skills and resource acquisition ability through a combination of group and individualised care. The programme consisted of the following 4 key components: client-centred goal-setting and monitoring; peer counselling; self-advocacy training; and the personal use of resource funds. It was based on the concepts of client-centred practice and empowerment (70). Research revealed that this consumer-driven programme had a positive impact on the participants’ symptom severity (23), quality of life (23, 24) and resource acquisition (25). Two other studies have shown that the most valuable
aspect of group interventions for CFS patients is the exchange of experiences among participants (18, 19).

4.7 Stage of Change Model
Not all patients are motivated to participate in non-pharmacological interventions, like patient-education and self-management programmes, or are able or ready to follow health recommendations. One model that has been used frequently over the last few decades to facilitate behaviour change with health-related conditions is the Stages of Change Model, also known as the transtheoretical model of behaviour change (71, 72). This model proposes that there are predictable and time-related stages of change. The extent to which people are motivated and ready to change depends on which stage of change an individual is in. Stages included in this model are (71, 72):

1. Precontemplation: Individuals are not intending to take action in the foreseeable future. Most people in this stage tend to be unaware that they have a problem or are uninformed or underinformed about the consequences of a given behaviour.
2. Contemplation: Individuals are intending to take action in the foreseeable future. They are aware that they have a problem and begin to think seriously about resolving it but have not yet made a commitment to take action in the near future.
3. Preparation: Individuals are intending to take action within the time frame of one month. He or she generally has a plan of action. It is these individuals who should be recruited for action-oriented treatment programs.
4. Action: Individuals have made specific, overt modifications in behaviour within the recent past.
5. Maintenance: Individuals have sustained overt changes over time. Maintenance is a difficult stage to achieve. This stage extends from 6 months to an indeterminate period past the initial action.
6. Termination: This stage occurs when the problem no longer presents any temptation and individuals have 100% self-efficacy. However, termination may never occur, only maintenance becomes less vigilant (65).

In health care, it is considered to be useful to stage the client’s intentions and behaviours for change as well as strategies that will enable completion of the specific stage (65).
## 5 Material and methods

Three populations were evaluated in this study. An overview of the participants and methods used in studies 1 to 4 is provided below:

<table>
<thead>
<tr>
<th>Paper</th>
<th>Participants</th>
<th>Methods</th>
<th>Analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Paper 1</td>
<td>Adults with mild to moderate CFS who participated in the patient education programme at Oslo University Hospital (n = 10)</td>
<td>Focus group interviews</td>
<td>Thematic</td>
</tr>
<tr>
<td>Paper 2</td>
<td>Adults with mild to moderate CFS who participated in the self-management programme in a town in southeast Norway (n = 6)</td>
<td>Focus group interviews</td>
<td>Thematic</td>
</tr>
<tr>
<td>Paper 3</td>
<td>None</td>
<td>Systematic review of qualitative studies</td>
<td>Meta-ethnography</td>
</tr>
<tr>
<td>Paper 4</td>
<td>Adults with mild to moderate CFS (n= 137) recruited from 4 towns in southern Norway and 2 suburbs of Oslo (n = 137); 118 participants completed all of the assessments (59 from each group)</td>
<td>Randomised controlled trial with two arms: 1) Intervention group: Participated in the self-management group 2) Control group: Received treatment as usual</td>
<td>A mixed-model analysis for repeated measures was performed to examine the differences in the primary and secondary outcomes. The paired-samples t-test was used for within-group comparisons.</td>
</tr>
</tbody>
</table>
5.1 Participants

5.1.1. Participants in the patient education programme (paper I)
Patients who participated in the CFS patient education programme at Oslo University Hospital from February to April 2010 were invited to participate in this study, which aimed to elicit participants’ experiences with the programme and their views regarding the usefulness of the programme. Nineteen of the 33 participants agreed to participate in the study. The inclusion criteria were meeting the CDC-1994 case definition (5) and the Canadian ME/CFS case definition (26) and an age of at least 18 years. Thirteen individuals were eligible for inclusion in the study. Three participants withdrew prior to the first interview. The final sample comprised eight women and two men. The participants’ mean age at the time of study inclusion was 43.7 years (range 32-57), and the mean duration of illness was 6.6 years (range 2.5-13.5). The sample represented a variety of social backgrounds. Only one participant was working, six participants had higher education, and six were married or lived with a partner.

5.1.2. Participants in the self-management programme (paper II)
The initial version of the self-management programme was developed in cooperation with an occupational therapist in primary healthcare (Live Lange Hellum) and a representative of the Norwegian ME Association (Hilde Hassum Aannestad). The programme feasibility, acceptability and beneficence were tested in 2011 in a pilot study set in a mid-sized town in southern Norway. The participants were required to have been diagnosed with CFS by a medical practitioner and to be at least 18 years of age. After the programme ended, all of the participants were invited to participate in focus group interviews to share their experiences. Six of them agreed to participate. The sample comprised five women and one man. The mean age at inclusion was 38.7 years (range 28-46), and the participants had been diagnosed with CFS 1 week to 7 years (mean 2.3 years) prior to their participation in the programme. The sample represented a variety of social backgrounds. Only one participant was working, four had higher education, and five were married or lived with a partner.

5.1.3 Participants in the randomised controlled trial (paper IV)
The final version of the self-management programme was evaluated in a randomised controlled trial. The participants in this study were recruited from November 2011 to January 2012 from four mid-sized towns in southern Norway and two suburbs of Oslo and their surrounding municipalities. The inclusion criteria were an age greater than 18 years, a diagnosis of CFS by a physician or medical specialist, meeting the CDC-1994 case definition.
(5) and the research or clinical ME/CFS Canadian case definition (26), not being pregnant, able to read and speak Norwegian, and considered physically able to attend the programme. A total of 146 participants were included in the study. These participants were randomly assigned to the intervention or the control group, each of which included 73 participants. A computerised randomisation list was created in blocks of six with the municipality as the stratifying variable (73). After randomisation, nine participants withdrew from the study; two withdrew from the intervention group because they were too ill to participate, and seven withdrew from the control group because they were too ill (1), because they did not accept their randomised group assignment (1) or for an unknown reason (5). A total of 137 subjects remained in the study at baseline, and 118 completed the assessments at one year. Therefore, the total dropout rate was 13.9 %. The participants’ baseline characteristics are presented in Table 5.1.

Table 5.1 Characteristics at baseline: mean (SD) or N (%)

<table>
<thead>
<tr>
<th></th>
<th>Intervention (N = 71)</th>
<th>Control (N = 66)</th>
<th>P-values</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>44.0 (11.8)</td>
<td>43.9 (11.6)</td>
<td>0.92</td>
</tr>
<tr>
<td>Diagnose (years)</td>
<td>Median: 3 years</td>
<td>Median: 3 years</td>
<td>0.69a</td>
</tr>
<tr>
<td>Female gender</td>
<td>67 (94.4)</td>
<td>54 (81.8)</td>
<td>0.022b</td>
</tr>
<tr>
<td>Living alone</td>
<td>19 (26.8)</td>
<td>14 (21.2)</td>
<td>0.45b</td>
</tr>
<tr>
<td>Children (number)</td>
<td>1.3 (1.2)</td>
<td>1.4 (1.2)</td>
<td>0.39</td>
</tr>
<tr>
<td>Has children under 18</td>
<td>30 (42.3)</td>
<td>30 (45.5)</td>
<td>0.71b</td>
</tr>
<tr>
<td>Education &gt; 12 years</td>
<td>38 (53.5)</td>
<td>31 (47.0)</td>
<td>0.44b</td>
</tr>
<tr>
<td>Workingc</td>
<td>3 (4.3)</td>
<td>8 (12.3)</td>
<td>0.09b</td>
</tr>
<tr>
<td>Student</td>
<td>2 (2.8)</td>
<td>0 (0.0)</td>
<td>0.17b</td>
</tr>
</tbody>
</table>

All analysed by independent-samples t-test except:

a Analysed by Mann-Whitney test
b Analysed by chi-square test
c1 missing in intervention and control group

Figure 5.1 shows the flow chart of the participants in study IV.
Figure 5.1 Flow chart of the participants in the RCT (paper IV)

- Interested after having received information (n = 213)
- Returned written informed consent (n = 180)
- Excluded (n = 34)
  - Not meeting inclusion criteria (n = 24)
  - Not receiving medical information (n = 10)
- Randomised (n = 146)
  - Allocated to intervention (n = 73)
    - Accepted participation (n = 71)
    - Follow-up after intervention (n = 67)
    - Follow-up at 6 months (n = 63)
    - Follow-up at 1 year (n = 59)
  - Allocated to control (n = 73)
    - Accepted participation (n = 66)
    - Follow-up after intervention (n = 64)
    - Follow-up at 6 months (n = 62)
    - Follow-up at 1 year (n = 59)

Intervention group: Lost to follow-up after accepted participation (n = 12) due to ill-health (1), moving to another town (1), and refused the evaluations (10)

Control group: Lost to follow-up after accepted participation (n = 7) due to ill-health (1) and refused the evaluations (6)
5.2 Methods

Complex interventions are those that contain several interacting components (74, 75). Self-management programmes are complex interventions because they include interacting components such as contexts, patients, programme leaders, patient education and self-management skills. It can be difficult and time-consuming to determine the effectiveness of complex interventions, especially when there is no adequate description of the development and content of the intervention (76, 77). Therefore, the Medical Research Council in the United Kingdom developed a framework for developing and evaluating complex interventions to improve health (76). This framework, which was updated and extended in 2008 (74), was developed to help researchers recognise and adopt appropriate methods. We were inspired by this framework when developing, piloting and evaluating the self-management programme in the present study. The key elements of the process are described in Figure 5.2 (74).

Figure 5.2 Key elements of the development and evaluation process (74)

The stages in the process were considered ongoing and flexible and did not follow a linear or cyclical sequence (74). Qualitative and quantitative methods can be considered complementary in medical research (74, 78, 79), and both methods were applied in this study. Figure 5.3 presents an overview of our application of this framework in the present study.
5.2.1 Development (paper I, II and III)
The following methods were used to develop the self-management programme: focus group interviews (paper I and II), a review of the evidence base (paper II), and a systematic review of the evidence base (paper III).
Prior to this study, the traditional patient education programme for CFS patients at Oslo University Hospital was developed, conducted, and adjusted several times by healthcare workers in co-operation with two peer counsellors (i.e., experienced CFS-patients). The programme that was evaluated in this study intended to promote coping by providing participants with information about different aspects of the illness. It included the following topics: coping skills, current medical approaches, personal relationships, pacing and energy conservation, physical exercise and relaxation, nutritional approaches, and economic self-sufficiency. The programme lecturers were experienced healthcare workers (a physician, occupational therapist, physiotherapist, psychiatric nurse, dietician, and a social worker) with competence in CFS. Each lecturer was responsible for teaching a topic during one of the programme meetings. A peer counsellor shared her own experiences with the illness and useful coping skills during one of the meetings and acted as a positive role model during the others meetings. During the final meeting, the lecturers and the peer counsellor answered the participants’ remaining questions. Some of the lecturers focused mainly on providing information, whereas others combined lectures with group discussions. The participants met eight times over a nine-week period. Each meeting lasted two hours, including a 30-minute break with the option to engage in relaxation exercises.

Focus group interviews were conducted to elicit the participants’ experiences with this patient education programme and their views regarding the usefulness of the programme. Focus groups were used because group dynamics among participants may generate a larger and richer range of ideas compared with individual interviews (80, 81). The focus group interviews were conducted immediately and nine months after participation in the programme. The semi-structured interview guide covered topics such as the perceived helpfulness of the different sessions, the perceived benefits of the whole programme, ideas about the illness, psychological coping processes, and applied coping strategies. The interviews lasted 95 – 110 minutes, and were audio-recorded and transcribed verbatim. The main investigator acted as the moderator and one of the co-authors was an observer during both interviews. The results of this study were used in the development of the self-management programme.

Because the framework of the Medical Research Council in the United Kingdom recommends considering the implementation of an intervention in the early stages of its development (74),
an occupational therapist in primary healthcare was involved in the entire process. In addition, the framework recommends that users be involved at all stages of the development process because they are likely to provide input that results in a better and more relevant intervention (74). Therefore, a representative of the Norwegian ME Association was involved in the entire process, and the participants were invited to share their experiences after taking part in the programme.

The occupational therapist, the Norwegian ME Association representative (a peer counsellor) and the main investigator held several meetings to develop the initial version of the programme. Their co-operation was based on co-production (53, 54), as defined in § 4.5.3. An essential early task was to develop a theoretical understanding of the likely change process by drawing on existing evidence and theory (74). The initial self-management programme was based on the concepts of client-centred practice, empowerment, self-efficacy and adult education, as presented in Chapter 4. In addition, the programme was based on previous research regarding patient education and self-management education, including study I. We also integrated clinical experiences with treating CFS patients in primary and specialised healthcare and personal experiences with CFS. The self-management programme that was developed in this study may therefore be considered an evidence based intervention, defined by Straus and colleagues (82, page 1) as ‘the integration of best research evidence with our clinical expertise and our patient’s unique values and circumstances’. The initial programme is presented in table 5.2.

| Eight biweekly sessions of 2.5 hours duration |
| Day 1: Information about the programme: content and form |
| Participants and leaders introduce themselves |
| Educational presentation about current medical approaches |
| Educational presentation about self-management |
| Selecting individual goals and constructing action plans to achieve these goals |
| Participants identify topics relevant for better coping with CFS and vote on the seven topics they consider as most relevant |
| Two participants take responsibility for preparing a short presentation or questions for dialog concerning each topic. They may involve one of the leaders or an external lecturer |
| Days 2-8: Participants present and evaluate their individual goals and action plans and receive feedback/problem solving |
| Educational presentation on one of the chosen topics by participants with exchange of individual experiences |
| Constructing action plans |

**Table 5.2 Content of the initial programme**
During the programme, coping capabilities were developed through the exchange of knowledge and experiences among participants, the modelling of self-management skills, guided mastery practice, and informative feedback (55). The programme covered a number of topics that were chosen by the participants. At the beginning of the programme, the participants set personal goals for coping and constructed action plans for achieving these goals. During each session, the action plans were evaluated, and the participants were offered feedback from other participants to help them overcome the obstacles they faced. At the end of each session, the action plans were adjusted or replaced with new action plans, as necessary. The initial programme was tested in the piloting stage, as described below (§ 5.2.2). The experiences of the group leaders and participants were used in the development of the final programme. A manual for programme leaders was developed prior to the piloting stage and adjusted prior to the evaluation stage.

Review and synthesis of qualitative studies concerning coping with CFS (paper III)

A systematic review of qualitative studies was performed to identify factors that promote coping in individuals with CFS and to provide recommendations for appropriate interventions for this patient group. Article selection began with a search for relevant studies that had been published before October 2013 using the following major scientific databases: PubMed, MEDLINE, PsycINFO, AMED, EMBASE, Web of Science and CINAHL. The databases were searched using the following subject headings and keywords. For population, we used the following terms: fatigue syndrome, chronic; chronic/postviral fatigue disorder/syndrome; encephalomyelitis, myalgic. For coping, we used the following terms: adaptation, psychological; coping; activities of daily living; quality of life; well-being; health knowledge/attitudes/practice; attitude to health; client attitudes; sick role; illness behaviour; coping behaviour; life experiences; emotional adjustment; self-concept; self-assessment; self-efficacy; patient satisfaction; client satisfaction; patient preferences; needs assessment; psychological needs. For methods, we used the following terms: qualitative research; interviews as topic; focus groups; narration; interview, psychological; grounded theory. In addition, three reviews associated with CFS (2, 11, 28) were screened to identify additional articles.

The study inclusion criteria were as follows: 1) the study was an empirical qualitative study; 2) all of the study’s subjects were diagnosed with CFS by a physician/specialist or met specified criteria for CFS; 3) the majority of the study participants were older than 18 years;
and 4) the studies focused on coping.

First, the studies that, broadly defined, used qualitative methods were identified. Second, the abstracts and titles were screened. Third, the potentially relevant articles were retrieved as full-text articles and assessed for inclusion using an include/exclude form. Finally, a quality assessment was performed according to a checklist for qualitative studies (78). The last three steps were performed independently by the main investigator and co-supervisor Elin Bolle Strand. Differences between the reviewers’ assessments were resolved via discussion. The synthesis of the qualitative studies was reported in accordance with the Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) framework (83). The results of study III were used to develop the final version of the self-management programme.

5.2.2 Feasibility/Piloting (paper II)

The initial version of the self-management programme was tested for its feasibility, acceptability and beneficence in the spring of 2011 in a pilot study that took place in a mid-sized town in southern Norway. Initially, the programme was led by the occupational therapist, the peer counsellor and the main investigator. However, by the end of the programme the main investigator acted primarily as an observer to determine whether the programme could be led by one occupational therapist and one peer counsellor, as intended.

Immediately after each session, the three programme leaders met to evaluate the content of the session, the group process, and our own performances as programme leaders, and we made the necessary adjustments to increase the feasibility of the programme.

After the participants completed the programme, they evaluated its feasibility. Again, focus group interviews were used. The first focus group interview occurred immediately after the programme ended. The semi-structured interview guide covered themes such as the perceived benefits of participating in the programme, the aspects of the programme that were experienced as helpful, the aspects that were experienced as less helpful, and suggestions for improving the programme. The results of the first focus group interview were validated by the participants during a second focus group interview conducted four months later. The proposed changes to the programme were discussed with the participants, and the participants were again asked about their perceived benefits from participating in the initial programme to provide insight into developments over time. The focus group interviews lasted 30-90 minutes. All of the interviews were recorded and transcribed verbatim. The main investigator
acted as the moderator. The occupational therapist was an observer during the first interview, and the occupational therapist and peer counsellor were observers during the second interview.

A power analysis, as presented in § 5.3.3, revealed that the evaluation stage had to include 112 participants. We expected a dropout rate of approximately 25% (42); therefore, we planned to include 150 participants. The recruiting process during the piloting stage provided indications of the size of the recruiting area and the number of locations needed for the RCT. The piloting stage also provided indications of the preferable group size per location.

The outcome measures were completed by and discussed with two patients to evaluate the feasibility of the outcome measures that were chosen for the RCT. The patients indicated that the chosen questionnaires did not require excessive effort or time to complete.

5.2.3 Evaluation (paper IV)
The results of the piloting stage of study II and the systematic review (study III) were used to develop the final programme (see Table 5.3).

<table>
<thead>
<tr>
<th>Table 5.3 Content of the final programme</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Eight biweekly sessions of 2.5 hours duration</strong></td>
</tr>
<tr>
<td><strong>Day 1:</strong> Information about the programme: content and form</td>
</tr>
<tr>
<td>Participants and leaders introduce themselves</td>
</tr>
<tr>
<td>Educational presentation about current medical approaches</td>
</tr>
<tr>
<td>Educational presentation about self-management</td>
</tr>
<tr>
<td>Discussing relevant individual goals</td>
</tr>
<tr>
<td><strong>Day 2:</strong> Educational presentation on one of the topics* with an exchange of individual experiences</td>
</tr>
<tr>
<td>Relaxation exercise</td>
</tr>
<tr>
<td>Selecting individual goals and constructing action plans to achieve these goals</td>
</tr>
<tr>
<td><strong>Days 3-8:</strong> Educational presentation on one of the topics* by the leaders with an exchange of individual experiences</td>
</tr>
<tr>
<td>Relaxation exercise</td>
</tr>
<tr>
<td>Participants present and evaluate their individual goals and action plans and receive feedback/problem solving</td>
</tr>
<tr>
<td>Constructing action plans</td>
</tr>
<tr>
<td>One meeting for relatives</td>
</tr>
</tbody>
</table>

*Topics days 2-8: Activity pacing and energy conservation, sleep and relaxation, physical exercise, nutritional approaches, economic self-sufficiency, personal relationships, and available treatments. Presentations are made by healthcare professionals from the CFS/ME-centre, Oslo University Hospital.
The final programme included eight fixed topics. Short presentations covering these topics were provided by professionals from the CFS/ME-centre. Compared with the initial version of the programme, the sequences of the course elements were changed; educational presentations on the topics with an exchange of personal experiences were scheduled prior to the break, while problem-solving and the development of actions plans were scheduled after the break. Additionally, relaxation exercises and a meeting for relatives were added.

A randomised controlled trial was used to evaluate the effectiveness of the self-management programme because randomised controlled trials provide the best evidence on the effectiveness of healthcare interventions (84). This trial comprised two arms: an intervention group that participated in the self-management programme and a control group that was offered a change to participate in the self-management programme after the study’s final outcome measures were completed. The control group was allowed to receive treatment as usual but was excluded from participating in the patient education programme at our hospital. The trial was reported in accordance with the CONSORT guidelines (85).

**Outcome measures**

We performed assessments at baseline and at six-month and one-year post-intervention. The one-year follow-up measurements were performed to evaluate whether the changes found at the six-month follow-up were sustained at the one-year follow-up.

A range of measures is needed to evaluate the effectiveness of complex interventions (74). The outcome measures were collected using questionnaires that the participants completed at home and returned by mail. The assessor was blinded to the group allocation of the participants.

**Physical functioning:** The Medical Outcomes Study 36-item Short Form Questionnaire (SF-36) is a health survey with scales that assess physical functioning, role-physical functioning, bodily pain, general health, vitality, social functioning, role-emotional functioning, and mental health (86). The physical functioning subscale served as the primary outcome measure (scores range from 0-100, with higher scores indicating better levels of functioning). This subscale has been used as a primary outcome measure in several other CFS studies (33, 42). The SF-36 has been validated in several international and national studies and has demonstrated adequate psychometric properties as a measure of functional status in a CFS population (87).

We also used some secondary outcome measures, as follows:

**Fatigue:** The fatigue severity scale (FSS) developed by Krupp and colleagues (88) was
used to measure fatigue severity, which is the main symptom of CFS (scores range from 9-63, with higher scores indicating more severe fatigue). The FSS has demonstrated adequate psychometric properties (89) and was found to be appropriate for measuring fatigue in people with CFS (90). This measure has been used in several CFS studies (42, 91).

**Self-efficacy:** The self-efficacy scale (92) was used to measure participants’ sense of control in relation to their CFS complaints. The ratings for each of the scale’s five questions are summed (the total score ranges from 5-24), and higher scores reflect higher levels of self-efficacy. The scale’s Cronbach’s alpha reliability has been shown to range from 0.70 to 0.77 (92). The scale has been used in several CFS studies (42, 92).

**Acceptance:** The illness cognition questionnaire (ICQ) measures illness cognitions in patients with chronic diseases (93). The ICQ consists of 18 items. For the current study, the acceptance subscale (six items) was used (scores range from 6-24, with higher scores indicating higher levels of acceptance). Research has indicated that the ICQ is a reliable and valid instrument (93). The ICQ has been used in CFS studies (94, 95).

**Health status:** The eight scales of the SF-36 (86) can be aggregated into two summary measures based on the weighted sums of the scale scores. The two summary measures are the physical component summary (PCS) for physical health and the mental component summary (MCS) for mental health (96). Because some discrepancies between the summary scores and the scores on the individual scales have been observed (97), oblique scores were used. The PCS and MCS oblique scores were computed using an online calculator provided by the SF-36 Community (http://www.sf-36.org), and the scores are based on Norwegian reference data. PCS and MCS scores below 40 points indicate poor health (98). The PCS and MCS have been used in CFS studies (99).

**Group leader training and monitoring**
The occupational therapists and peer counsellors who led the self-management programme participated in a three-day training seminar that focused on imparting knowledge about CFS/ME, the content of the programme, and handling group-processes. A manual for programme leaders was developed to ensure that each group intervention was delivered using the same approach. Monitoring occurred via telephone and addressed specific problems that the programme leaders experienced during the sessions. The programme leaders were also invited to meet immediately after the intervention to evaluate the self-management programme.
5.2.4 Implementation

The evaluation phase revealed that longer-term quantitative and qualitative follow-up studies are needed before firm conclusions can be drawn regarding the effectiveness of the self-management programme. In addition, further development of the programme followed by piloting and evaluation may be needed before the programme can be implemented in a primary healthcare setting. Further development of the programme may also require revising the manual and training programme for future programme leaders. Future applications of the programme must include planning for surveillance and monitoring before the programme is implemented.

5.3 Analysis

Given the use of different approaches, a variety of analyses were used.

5.3.1 Qualitative analysis (paper I and II)

Thematic analysis of the focus group interviews in these studies followed Richards and Morse’s (100) description and involved a series of steps. First, the interviews were read thoroughly to form a general impression of their content. Second, topic coding was used to identify and label sentences and paragraphs on a topic or theme. Third, topics that appeared to pertain to a similar phenomenon were compared and related to one another. Finally, abstraction was achieved by gathering topics in categories.

5.3.2 Synthesis of qualitative studies (paper III)

Different methodologies have been presented for synthesising qualitative studies (101). Meta-ethnography (102) is perhaps the most well developed method; therefore, it was selected for this study. The meta-ethnographic synthesis of interpretive research involves translating the studies into one another; the interpretations and explanations in the original studies are translated across the studies to produce a synthesis (102). Noblit and Hare (102) outlined a series of seven phases involved in performing a meta-ethnography (Box 5.1).
Box 5.1 The seven phases of Noblit & Hare’s meta-ethnography

1. Getting started
2. Deciding what is relevant to the initial interest
3. Reading the studies
4. Determining how the studies are related
5. Translating the studies into one another
6. Synthesising translations
7. Expressing the synthesis

The analysis began with repeated readings of the articles. In the next phase, we determined how the studies were related. We examined the articles and created a list of the reported coping concepts using the terminology from the original articles. Next, these concepts were aggregated under the emerging key concepts. By reviewing the list of concepts and key concepts, we were able to establish the relationship between the studies and aggregate them in an overarching translation (second-order interpretations).

5.3.3 Quantitative approaches (paper IV)

Power analysis
In a previous publication (103), we found that to obtain 80 % power with a 5 % significance level, a minimum of 56 participants must be included in each group to detect a 15-point difference between the groups (SD = 28) on the SF-36 physical functioning subscale. We expected a dropout rate of approximately 25 % (42); therefore, we planned to include 75 participants in each group.

Statistical analysis
SPSS, version 15 (SPSS Inc., Chicago, IL, USA) was used for the statistical analyses. At baseline, the independent-samples t-test was used to compare the primary and secondary outcomes between the groups. In addition, the Mann-Whitney U-test was used to compare continuous variables that had a skewed distribution, whereas the chi-squared test was used to compare binary variables. The paired-samples t-test was used for within-group comparisons between baseline and the six-month and one-year follow-ups. Analyses were conducted as ‘per-protocol’ analyses, in which only the participants with complete registrations for an outcome measure were included in the statistical analyses of that measure. A significance level of 5 % was used.
To investigate the effect of the intervention on physical functioning, fatigue severity, self-efficacy, acceptance and health status compared with controls, a mixed-model analysis for repeated measures was performed. The time of measurement (baseline and six-month and one-year follow-ups) was used as a within-subjects factor, and the group (intervention or control) was used as a between-subjects factor.

When less than 50 % of the values on a subscale were missing, the values were replaced using an imputation procedure in SPSS. A total of 134 (0.003 %) of 47,012 possible values were imputed.

5.4 Ethical considerations
The first study was approved by the Regional Committee for Medical Research Ethics (Health Region South-East, approval number 2009/755/REK sør-øst). The second and fourth study were approved by the Regional Committee for Medical Research Ethics (Health Region North, approval number 2011/894/REK nord). In addition, the studies were approved by the Privacy Ombudsman for Research at our hospital. The fourth study was also registered in Clinical Trials.gov, identifier: 2011/894/REK nord.

Written informed consent was obtained from all of the participants in studies I, II and IV following a written and oral explanation of the study. Participation in these studies was voluntary, and the participants were informed of their right to withdraw from the study at any time without further explanation. To protect the anonymity of the participants, information that might lead to personal identification was removed from the papers.

We expected that participation in the focus group interviews (studies I and II) and the programme (studies I, II, and III), including transportation, might incidentally lead to increased tiredness and fatigue. Therefore, the focus group interviews lasted no more than approximately one to two hours, whereas the group sessions in studies II and IV lasted 2.5 hours, which included a 30-minute break. If the participants felt tired during the focus group interviews or the sessions, they were invited to rest or, in a few cases, advised to withdraw from further participation in the study.

In study IV, the participants completed the questionnaires at home and returned them via mail. Calculations determined that it would take approximately 30 to 60 minutes to complete all of the forms. To avoid over-exertion, the participants were given several weeks to complete the questionnaires. In addition, they were encouraged to accept help with completing the questionnaires, including help from a research assistant.

During the intervention, the programme leaders and participants established a good
relationship. Most of the participants reported that their experiences with participating in the programme were positive. Occasionally, the participants reported major problems related to their everyday life and/or psychological well-being. Such situations were handled by encouraging the participants to contact the appropriate healthcare professionals and supporting their efforts to do so.
6. Results – abstracts of the papers

*Paper I*

**Title:** From chaos and insecurity to understanding and coping: Experienced benefits of a group-based education programme for people with chronic fatigue syndrome.

**Objective:** The aim of this study was to elicit participants’ experiences with a multidisciplinary patient education programme and their views regarding the usefulness of the programme.

**Methods:** Focus group interviews were conducted with ten participants immediately and nine months following participation in the programme and were analysed using thematic analysis.

**Results:** Initially, the participants experienced confusion and insecurity regarding coping with their illness. Participation in the programme appeared to promote understanding, acceptance and coping through gaining greater knowledge, learning coping strategies, exchanging experiences, as well as receiving understanding and acceptance.

**Conclusions:** The programme was experienced to be a beneficial intervention. However, the participants expressed a need for more guidance or follow-up to maintain the practice of coping strategies for better coping with their illness.
Title: Development of a group-based self-management programme for individuals with chronic fatigue syndrome: A pilot study.

Objective: The aim of the study was to develop a group-based self-management programme for individuals with chronic fatigue syndrome (CFS) by using the participants’ experiences with the initial version of the programme, which intends to promote coping with the illness in a primary healthcare setting.

Methods: We developed an initial programme, based on self-efficacy theory and the concepts of client-centred practice and empowerment. Subsequently, the programme was tested and further developed by drawing on the participants’ experiences with the programme. Focus group interviews were applied. The interviews were analysed using thematic analysis.

Results: The initial programme was determined to be feasible, although several modifications regarding the content and the practical organisation of the programme were proposed.

Conclusion: In line with the participants’ experiences, we developed the final self-management programme, which includes short presentations of eight topics, the exchange of experiences among participants, goal-setting, the construction of action plans, and relaxation exercises, in addition to a meeting for relatives. The programme will be provided in eight biweekly sessions and will be led by juxtaposed peer counsellors and occupational therapists. The effects of the final programme will be evaluated in a randomised controlled trial.
Title: Coping with chronic fatigue syndrome: A review and synthesis of qualitative studies.

Objectives: The objective of this study was to identify factors that promote coping with chronic fatigue syndrome (CFS) and to provide recommendations for adequate interventions in this patient group.

Methods: We performed a systematic review of qualitative studies that focused on coping with CFS. Structured searches were conducted in major scientific databases. Two reviewers independently performed the study selection and assessment. Meta-ethnography was applied to review and synthesise the studies. The analysis provided key concepts that could be aggregated using second-order interpretations.

Results: Fifteen studies were included. The findings suggest that coping strategies, including activity management and the use of cognitive and emotional strategies, and psychological processes such as acceptance and the rebuilding of identities and lives, may promote coping with the illness. The use of adequate coping strategies appeared to be facilitated by progress in psychological processes. Coping with CFS appeared to be facilitated mainly by self-management, occasionally complemented by treatment and social support.

Conclusions: The findings indicate that interventions that focus on coping with CFS may include the use of adequate coping strategies and may take into account progress in psychological processes. Interventions may be complemented by self-management, other types of treatments, and social support.
Title: Effectiveness of a group-based self-management program for people with chronic fatigue syndrome: A randomized controlled trial.

Objective: The object of this study was to evaluate the effectiveness of a self-management programme for people with chronic fatigue syndrome (CFS) in a primary healthcare setting.

Methods: We conducted a randomised controlled trial with 137 participants, assessing physical functioning, fatigue severity, self-efficacy, health status and acceptance at baseline, and at six-month and one-year post-intervention.

Results: No significant differences were found within or between the groups in physical functioning. At the six-month follow-up, a significant difference between the two groups was found concerning fatigue severity ($p = 0.039$) in favour of the control group, and concerning self-efficacy in favour of the intervention group ($p = 0.039$). These significant differences were not sustained at the one-year follow-up. Acceptance improved significantly in the intervention group at the six-month (mean change = 0.9, $p = 0.006$) and one-year (mean change = 0.7, $p = 0.030$) follow-ups and in the control group at the six-month follow-up (mean change = 1.1, $p = 0.001$). The dropout rate was low (13.9 %) and the median number of sessions attended was high (seven of maximum eight).

Conclusions: Long-term follow-up studies are needed before firm conclusions can be drawn on the effectiveness of the intervention.
7. Discussion
The overall aim of this study was to develop and evaluate a group-based self-management programme for individuals with CFS that promotes coping with the illness and that can be implemented in a primary healthcare setting in Norway. The rationale for this study was based on several studies showing that patients receive insufficient information, support and help from healthcare professionals to cope with their illness. Our study is a contribution to address this paucity by undertaking the development, piloting and evaluation of a self-management programme for individuals with CFS.

The main findings will be discussed relative to the overall aim of this study and to the applied study design and methods.

7.1 Development of the programme
The results of studies I, II and III were used to develop the programme that was evaluated in study IV.

7.1.1 Understanding, acceptance and coping
The first three studies showed that coping appears to be facilitated by progress in psychological processes such as acceptance (study I, II and III) and the rebuilding of identities and lives (study III). Moreover, in study III, we emphasised that interventions must focus on psychological processes to promote coping.

Several other studies concerning CFS have revealed that understanding and accepting the illness seems to facilitate coping (1, 104) because acceptance may promote the downgrading of expectations and the search for new sources of fulfilment (1, 11, 104). Studies I and II revealed that acceptance was promoted by the new insights and understandings that were gained through participation in the patient education programme (study I) and the self-management programme (study II). In both studies, new insights and understandings were gained through the assimilation of knowledge about the illness and coping strategies and through the exchange of experiences with fellow participants.

7.1.2 Knowledge about the illness and coping strategies
The programme in study I encompassed eight fixed topics illuminating different aspects of the illness, while the topics in study II were chosen by the participants. The participants in study II initially appreciated being empowered to choose the topics that the programme would cover. However, they concluded that the final programme did not need to include the opportunity to choose topics when they learned that the topics they had chosen were similar to
those covered in study I. The chosen topics were also similar to those that were covered in the CFS empowerment project (25) and those that were found in the systematic review (study III). These similarities may indicate that these topics are the ones of greatest concern to CFS patients, which was why they were included in the final programme that was evaluated in study IV. However, new knowledge about CFS may modify the needs of CFS-patients over time and may entail the need to evaluate and adapt the programme content regularly.

All three studies revealed that pacing of activities appeared to be an important coping strategy. The usefulness of pacing, however, is disputed among scientists because quantitative studies have shown conflicting results for interventions that include (33, 39). Slightly divergent interpretations of pacing in these studies may explain the differences in outcomes (37) because practising pacing by under-expending available energy may not be beneficial (105). However, several qualitative studies support the findings of our study (2, 11, 106, 107).

Our findings are also consistent with the results of a survey sponsored by the ME Association in the United Kingdom (32). Therefore, we decided to include pacing as one of the topics in the final programme.

In studies I and III, we found that the use of relaxation and meditation techniques may reduce stress levels and facilitate better rest. Other studies have shown that such techniques can have a positive effect on symptoms (42) and may improve patients’ mood, quality of life and physical functioning (108). Therefore, we decided to add relaxation exercises to the final programme.

**7.1.3 Meeting fellow participants**

The participants in both studies I and II reported that meeting fellow patients was helpful; for some participants, this was the most valuable part of the programme. The participants in both studies appreciated exchanging coping experiences and beneficial coping strategies. These findings were consistent with the results of two others qualitative studies concerning group interventions for CFS patients (18, 19). In addition, the participants in study I had an overall positive experience regarding the understanding and acceptance provided by fellow participants who were experiencing the same types of symptoms and problems. The participants felt that this mutual understanding created a safe environment for discussing issues that the participants had not been able to discuss elsewhere. These results emphasise the need to allow sufficient time for group discussions in the final programme.

Several other studies have revealed that sufferers desire fellowship with other CFS patients to receive support and share experiences and information (29, 109-111), e.g., via
support groups and online. Åsbring and Närvänen (29) observed that this sharing is not necessarily a positive experience for all patients. One reason that sharing was a positive experience in studies I and II may be that the lecturers and programme leaders focused on constructive solutions more than on negative experiences.

7.1.4 Goals and action plans
The programme that was used in study I could be considered a traditional patient education, whereas the programme in study II combined traditional patient education with self-management education. The participants in both studies reported improved understanding, acceptance and coping as a result of the programme, especially at the follow-ups. However, these benefits were reported at the nine-month follow-up in study I and at the four-month follow-up in study II. Although we cannot rule out the possibility that the participants in study I might have reported the same results after four months, the difference might be explained by the use of goals and action plans in study II. Study III revealed the importance of regaining personal agency and taking control to identify methods for coping with the illness; however, the participants in several of the included studies reported that it was impossible to gain full control remained impossible because of problems associated with putting advice and knowledge into practice, among other reasons. As mentioned in § 4.5.9, setting realistic goals serves as a motivating factor that may promote learning and health behaviour (55, 65). Study III confirmed these assertions. Moreover, the use of action plans to achieve goals makes it possible to find solutions and learn problem-solving skills that may promote coping with chronic illness (22). Several studies have shown that programmes that include self-management action plans are more effective at improving outcomes than are programmes that do not include action plans (22). Goal-setting and action planning were therefore included in the final programme.

7.1.5 Programme lecturers
In study I, information was provided by experienced healthcare professionals with competence in CFS and by one peer counsellor; in study II information-giving was based on the exchange of knowledge among participants through presentations and group discussions. This difference did not seem to influence the participants’ perceptions of the programme’s benefits. The participants in study II, however, felt unsure about the extent they could trust information they had found out by themselves. In addition, the participants reported that preparing presentations was excessively demanding. This is not a surprising result, as CFS is characterised by chronic fatigue and cognitive impairments (5, 26). These findings may
indicate that lectures should be delivered by healthcare professionals. Given the limited access to healthcare professionals with competence in CFS, however, the participants in study II proposed that short presentations covering the eight fixed topics could be made by professionals at the CFS/ME-centre and be included in the final programme.

The patient education programme in study I focused mainly on providing information and involved several healthcare professionals. In contrast, in study II, less time was devoted to providing information, allowing more time for group discussions and the assimilation of self-management skills. This approach involved only one healthcare professional with competence in CFS and one peer counsellor. The content and organisation of this programme made it possible to conduct the programme in a primary healthcare setting.

Many CFS patients experience difficulties with having their diagnosis properly acknowledged and understood by healthcare professionals (2). Consequently, the participants in study II appreciated that the programme was co-led by healthcare professionals with competence in CFS. However, it may be an advantage to include occupational therapists as programme leaders because several of the topics address issues that may be included in occupational therapy interventions, such as activity pacing, energy conservation, physical exercise and available treatments. Moreover, study II confirmed that self-management behaviour can be modelled by having peer counsellors teach self-management programmes (59). Several studies have shown that, when they are well trained and are given a detailed manual, peers can teach as well as healthcare professionals (67, 112). Thus, we decided that the final programme would be led by juxtaposed occupational therapists and peer counsellors. As recommended in study III, the programme leaders in study IV were trained to understand, recognise and validate CFS and to offer the participants emotional support and appropriate challenges.

7.1.6 Meeting for relatives

Patients with CFS often report that they are not understood by others, an issue that may lead to withdrawal from social life and a loss of friendships (1, 11, 29). The participants in study II expressed that their relatives needed information to help them better understand the illness. They acknowledged that children also needed information about CFS, but felt that a meeting for adults should be prioritised. Consequently, a meeting for relatives older than 18 year was added to the final programme.
7.1.7 Follow-up
Studies I and III revealed that coping with CFS appeared to be facilitated mainly by self-help that was complemented occasionally by treatment. The participants in study I reported that they lacked information about their illness and how to cope with it and that before they participated in the patient education programme, they had felt that they were left to their illness alone. Therefore, it may not be surprising that many CFS patients participate in support groups (113) and online forums (111). Studies I and II showed that participation in a patient education programme or self-management programme met the need for information about the illness and how to cope with it. They described being in the action stage of the Stages of Change Model (71, 72). But after participating in the programme, the participants in study I felt the need for additional guidance and follow-up to maintain the use of the coping strategies they had learned. Falvo (45) acknowledges that sustaining change may be difficult and individuals may struggle to prevent relapse. Continued support, encouragement, and reinforcements may help patients to maintain the changes they have made (45). After nine months, several participants had tried to obtain help but had difficulty finding primary healthcare professionals with competence in CFS.

In study II, the participants were encouraged to contact one another for support during the course of the programme. After they finished the programme, the participants formed a peer network, and approximately ten participants met once a month to socialise, provide support and exchange information and experiences. They did not report a need for follow-up from healthcare professionals. This result may indicate that the self-management skills that were developed and tested in study II allowed the participants to facilitate their own form of follow-up in a way that the participants in the patient education programme (study I) were unable to do. Consequently, the part of the programme that encouraged mutual support remained unchanged in the final programme.

7.1.8 Final programme
Based on the results of studies I, II and III, we developed the final programme (see Table 5.3), which consisted of eight biweekly sessions lasting 2.5 hours each. The programme combined patient education with self-management education. The programme was based on the concepts of client-centred practice (e.g., partnership, communication, goal-setting and respecting the participants’ ability to make choices and decisions) and empowerment (e.g., providing knowledge and establishing peer networks). In addition, the programme was based on self-efficacy theory (55), including guided mastery practice through goal-setting and
action-planning, modelling of self-management skills, providing informative feedback, and offering knowledge. The development of the programme was also guided by the six core learning assumptions about adult learners described by Knowles, Holton, and Swanson (60) and presented in § 4.5.6. The results of studies I and II showed that group education was perceived to be beneficial. The specific needs of individuals (63) were covered in group-discussions and in individual goal-setting and action planning. The selection of the learning activities and teaching tools used in the programme was guided mainly by the audience’s characteristics (i.e., adults with CFS), the learning objectives (coping with CFS) and cost effectiveness (62). The following instructional methods were used in the programme: lectures, group-discussions, role playing, role modelling, demonstration, one-to-one instruction, and self-instruction. The teaching tools that were used included PowerPoint presentations and printed materials (hand-outs and texts covering the presented topics and forms for goal-setting and action planning). The programme was led by juxtaposed peer counsellors and occupational therapists, who participated in a training programme prior to conducting the programme.

Based on the results of study II, the final programme included short presentations covering eight fixed topics made by healthcare professionals from the CFS/ME-centre. These presentations provided ‘quality assurance’ for the imparted information and concomitantly allowed sufficient time for the participants to exchange information and experiences that met their particular needs (63). This change in the programme may have strengthened the client-centeredness of the intervention because it may have met the needs of the participants in a better way and enabled them to make more informed choices (47). In addition, the inclusion of presentations made by healthcare professionals made it easier to include the three sets of self-management tasks delineated by Corbin and Straus (58) and the results of the review (study III).

Information about pacing was included in the programme. As mentioned in § 7.1.2, research has shown that slightly divergent interpretations of pacing may lead to differences in outcomes (37). Pacing, as presented in the programme, was based on the Energy Envelope Theory, which hypothesises that avoiding over-exertion by maintaining expended energy levels within the envelope of perceived energy levels might prevent relapses and increase energy levels and the tolerance for activity (30). The results of several studies of CFS patients support the value of the Energy Envelope Theory (25, 39).

Based on the results of study III, relaxation exercises were added to the final programme. A meeting for relatives was also added, as mentioned in § 7.1.6.
7.2 Evaluation of the programme
The final programme was evaluated in study IV. We formulated two hypotheses. Our primary hypothesis was that the intervention would lead to improvements in physical functioning for people with CFS compared with people with CFS receiving the usual care. The secondary hypothesis was that the intervention would lead to improvements in fatigue severity, self-efficacy, acceptance, and health status.

The trial revealed significant differences between the two groups concerning fatigue severity (in favour of the control group) and self-efficacy (in favour of the intervention group) at the six-month follow-up. These significant differences were not sustained at the one-year follow-up. The intervention had no effect on physical functioning, health status or acceptance for the participants with CFS compared with people with CFS who received the usual care.

Because pacing in our programme was based on the Energy Envelope Theory, we expected that participation in the programme would lead to improvements in physical functioning (39, 114); however, the intervention group reported no change in physical functioning. The lack of improvement in physical functioning may be explained by the finding that the majority of the participants in our study were already able to stay within their ‘energy envelope’ at baseline (Table 1) and, consequently, they might have experienced improvements in physical functioning prior to their participation in the programme. However, Brown and colleagues (105) determined that the patients who were closer to remaining within their ‘energy envelope’ demonstrated greater impairments in physical functioning compared with the patients who were moderately over-extending. This result may indicate that the use of more adaptive strategies is not associated with improvements in physical functioning (105).

Our programme encouraged the participants to increase their activity levels once they were able to manage their daily activities and their fluctuations in symptoms were reduced to a manageable level. The lack of improvement in physical functioning in the intervention group may indicate that the participants continued to operate at lower-than-recommended levels. The use of an objective measure of activity might have confirmed their compliance.

Based on the results of other studies that included pacing according to the Energy Envelope Theory, we also expected that the programme would lead to improvements in fatigue (39, 114). Throughout the entire study, the intervention group reported no change in fatigue severity, whereas the control group reported an improvement in fatigue severity. A significant difference between the two groups in favour of the control group was found at the six-month follow-up. This result is surprising and may indicate that remaining within the ‘energy envelope’ is not associated with improvements in fatigue either, as found for physical
functioning (105).

Similar studies have demonstrated improvements in self-efficacy (41, 42, 67, 69); however, we did not find improvements in self-efficacy in the intervention group. Moreover, it was surprising that the control group showed a significant decline in self-efficacy at the six-month follow-up. In addition, we found a significant difference between the groups at the six-month follow-up in favour of the intervention group, which indicated that participation in the self-management programme might have prevented a decline in self-efficacy. This result may indicate that the individuals in the control group did not receive adequate help and support from health professionals to enable them to cope with their illness (11). The finding that the difference between groups was not sustained at the one-year follow-up may indicate a need for follow-up over a longer time period (45, 65, 71, 72), similar to what the CFS Empowerment Project offered (23). Several studies have found an association between the acquisition of knowledge and improved understanding and acceptance (28). Because the exchange of information and experiences was part of the intervention programme, the intervention group showed significant improvements in acceptance at the six-month and one-year follow-ups, as expected. This result is positive because studies of CFS have revealed that accepting the illness seems to facilitate coping (1, 104). However, it was surprising that we did not detect differences in acceptance between the groups.

Van Damme and colleagues (95) found a positive relationship between acceptance and psychological aspects of well-being. In our study, however, both groups reported no change in their health status, which indicated that changes in health status do not necessarily occur immediately after improvements in acceptance.

This study had several limitations. It should be noted that our programme lasted for only 15 weeks, a duration that was based partly on the experiences with self-management programmes designed for people with chronic illnesses (67). Participants in other trials that examined CFS were seen more often and were followed for a longer period (23, 33, 42). Whiting and colleagues (115) determined that CFS studies that included a longer treatment duration were more likely to report that the intervention had positive effects, which may explain the lack of improvements observed in our study in terms of physical functioning and health status.

In addition, studies have indicated that learning that takes place during a group intervention must be integrated over time (116). Therefore, longer-term follow-up studies may be needed before firm conclusions can be drawn about the effectiveness of the programme.

Our programme included group sessions alone, whereas other interventions for CFS
typically include only individual consultations (33, 42) or a combination of group interventions and individual consultations (23). Our group intervention might have achieved better outcomes if we had also included individual consultations (55).

Individuals with CFS are a heterogeneous group; therefore, the evaluation of subgroups of individuals has been recommended. In our study, it would have been interesting to evaluate whether the self-management programme was more effective for patients who were over-extending. However, because of the relatively small sample size, it was not appropriate to classify individuals into subgroups according to their particular clusters of symptoms was not appropriate.

The low dropout rate and the high number of sessions attended may indicate that this programme is an acceptable and helpful intervention. This conclusion may raise the question of the extent to which the outcome measures that were applied in this study could detect the effects of the intervention.

Our results were obtained from a large randomised controlled trial with a low dropout rate (42) and a final measurement point at the one-year follow-up. This study design strengthens the reliability of our results.

In conclusion, the results of the present study suggest that the evaluated self-management programme for people with CFS had a stabilising effect on fatigue severity and self-efficacy for people with CFS at the six-month follow-up compared with people with CFS who received the usual care. Longer-term quantitative and qualitative follow-up studies are needed before we can draw firm conclusions about the effectiveness of our self-management programme. In addition, further development of the programme, followed by piloting and evaluation, may be needed before the programme can be implemented in a primary healthcare setting.

7.3 Evaluation of the study design and methods

One strength of the present study was that we followed the recommended guidelines for developing and evaluating a complex intervention for adults with CFS (74, 76) to provide transparent information about the study’s methodology and findings. In accordance with these guidelines, different methods were applied, including focus group interviews (papers I and II), a systematic review (paper III), and a randomised controlled trial (paper IV). Ideally, the systematic review of qualitative studies should be performed prior to the development of the initial programme, but because of time constraints, we performed the systematic review after the initial programme was tested in a pilot study.
7.3.1 Participant recruitment
The participants in study I were recruited from individuals who participated in the CFS patient education programme in the spring of 2010; only ten of the 33 participants participated in our study. Thus, the study population may not be representative of the total population that usually participates in our patient education programme. However, the sample represented variations in gender, illness duration, and social background. Moreover, the baseline scores on the questionnaires suggested that the participants were comparable to participants in similar studies (33, 117, 118).

The participants in studies II and IV were recruited from the town and suburbs where the programme was conducted and from the surrounding municipalities. The subjects were recruited from a variety of sources, including healthcare professionals, waiting lists for the CFS patient education programme at Oslo University Hospital, and CFS patient organisations. Information about the study was disseminated through brochures and personal communication that focused on recruiting participants that had terminated the precontemplation and contemplation stages of the Stages of Change Model (72). In addition, study announcements seeking participants were placed on the website of our hospital. The studies may still have been biased if we were unable to reach out to all potential participants with information about the study. The populations of studies II and IV may therefore not be representative of all individuals with mild to moderate CFS in Norway.

Moreover, only six of the 14 participants that finished the programme in study II were willing to participate in the focus group interviews. Thus, the responses obtained from these focus group interviews may not be representative of all of the participants. However, the information obtained through the focus group interviews was largely congruent with the experiences of the programme leaders.

7.3.2 Initial screening
For inclusion in studies I and IV, we used the CDC-1994 (5) case definition because it is frequently applied in research and because it is the most extensively validated case definition (6). However, research has shown that fatigue, post-exertional malaise, neurocognitive problems, and unrefreshing sleep are the most common symptoms reported by patients with CFS (119, 120). Some studies have shown that the Canadian ME/CF case definition (26) may be more sensitive than the CDC-1994 case definition (5) for identifying CFS-cases (121, 122). Therefore, the Canadian ME/CFS case-definition was also applied in studies I and IV. In addition, to exclude self-diagnosed participants, we requested confirmation of the
participants’ diagnoses from their GP’s or medical specialists.

In study I, a physician conducted the initial screening based on these criteria. In study IV, we did not have access to a physician. Therefore, the patients who were interested in participating in this study were asked to complete the DePaul symptom questionnaire (DSQ) (119, 123, 124). The DSQ is a self-report measure that contains items that measure the dimensions of the CDC-1994 (5) and the Canadian ME/CFS (26) case definitions by rating the frequency and severity of 54 symptoms over the prior six months on a five-point scale. The DSQ has been shown to have good test-retest reliability among patient and control groups (119) and good convergent and discriminant validity (124).

Because of time constraints, patients were included in study II only when they could confirm that they had been diagnosed with CFS by their GP or medical specialist. In this way, we excluded self-diagnosed patients; however, a variety of diagnostic criteria for CFS may have been used, resulting in substantial differences in the patterns of symptoms and disability among the included participants (125). The sample in study II may therefore be less homogenous than the samples in studies I and IV, and this heterogeneity may have influenced the findings of this study (125).

7.3.3 Focus groups (paper I and II)
In studies I and II, focus group interviews were conducted to elicit the participants’ experiences with the conducted programmes and their views about the usefulness of the programmes. Focus groups were used because the group dynamics among participants may generate a larger and richer range of ideas than individual interviews would (80, 81). A major limitation of the focus group method is the risk that certain participants will dominate discussion and exclude the contributions of others (80, 126). Therefore, the moderator has the important role of encouraging discussion and participation and refraining from giving preference to the speakers whose views are perceived as the most interesting (80).

Qualitative research is contextual and subjective, whereas quantitative research is generalisable and objective (127). Consequently, judgements about the quality of qualitative research in terms of reliability and validity (such as those used in quantitative research), are subject to debate (100, 128-130). One group argues that qualitative research must be judged according to the same criteria as quantitative research; a second group believes that a different set of criteria is required; whereas a third group of authors has questioned the appropriateness of any predetermined criteria for judging qualitative research (128). Most qualitative researchers seem to belong to the second group (128). Richards and Morse (100), for instance,
state that reliability requires that the same results are obtained if the study is replicated, and validity requires that the results accurately reflect the studied phenomenon. Moreover, Whittemore and colleagues (127) specify that the validity standards used in qualitative research must incorporate rigor and subjectivity as well as creativity.

In 1985, Lincoln and Guba recommended that the concepts of reliability and validity be translated into the following aspects of trustworthiness: internal validity into *credibility*, external validity into *transferability*, reliability into *dependability*, and objectivity into *confirmability* (131). Although not all qualitative researchers support these translated criteria, they seem to remain the gold standard (100, 127, 128).

To ensure *credibility*, a conscious effort must be made to establish confidence in an accurate interpretation of the meaning of the data (127, 131). This can be achieved by asking participants to confirm the results (i.e., member checking) (129, 131, 132). In studies I and II, member checking was performed in real time while the focus groups were conducted, meaning that the discussions about each identified issue were summarised and presented to the participants for confirmation or clarification before the researchers left one topic and continued the interview with another (132). Reconvening a focus group for member checking at a subsequent timepoint is often impractical, and the group dynamics will not be the same (132). Nevertheless, the results of the first focus group interview in study II were member checked during the second focus group interview because they formed the starting point for the second interview.

Authenticity is closely linked to credibility and refers to reflections on the meanings and experiences that are lived and perceived by the participants (127). Consequently, differences in perceptions, opinions and experiences must be reflected to secure authenticity (127). In addition, the researcher must evaluate whether any apparent agreement resulted from the coercion or self-censoring of participants with alternative viewpoints (132). In study I, the main investigator had two individual consultations with some of the participants prior to the patient education programme and was involved in the programme as a teacher during one of the eight meetings. In study II, three of the authors were both programme leaders and were involved in the focus group interviews as moderators or observers. These limitations may have influenced the extent to which the participants felt free to express their negative experiences.

Qualitative research requires an accurate description of the participants’ experiences. Therefore, the participants were informed that their anonymity was assured and that participation in the study would not have consequences for future treatment at our hospital. In
addition, the relationship between the researcher and the participants can become pseudo-therapeutic, and the researcher may have difficulties separating his or her own experiences from those of the participants, which may result in subjectivity (133). Therefore, the first author had to make an effort to focus solely on the experiences of the participants to produce theoretical understanding and sensitivity (133). In this way, she was able to construct meaning that would not be apparent to an external moderator. Despite these limitations, the focus groups in studies I and II displayed apparent polarisation that secured authenticity. However, confidence in the focus group findings can almost always be enhanced by including other data sources (132). Consequently, for a more detailed exploration of the observed differences, the authenticity of our studies could have been reinforced by adding individual interviews that were conducted by an independent investigator.

The distinction between internal and external validity may hold less meaning in qualitative research compared with quantitative research because generalisability to other populations is not a significant research goal (127, 131). To promote transferability, we made an effort to provide a thick description of the results of studies I and II to enable someone interested in making a transfer to reach a conclusion about whether a transfer is possible (131). Additionally, the results of these studies were compared with the results of other studies about coping with CFS (129, 132). The majority of our results were consistent with the results of these other studies. In addition, the baseline scores of the participants in study I were comparable to those of the participants in similar studies. The results of studies I and II may therefore be transferable to other people with mild to moderate CFS.

Given the complexity and lack of homogeneity of the focus group interviews, it may not be reasonable to assume that one rater can independently reproduce all of the coding for a given statement (132). Consequently, dependability was pursued by constraining the testing of agreement with the co-authors to core concepts, themes and categories (132). The internal consistency of the coding was enhanced by giving the main investigator the primary responsibility for conducting all of the analysis in study I and II and by regular communication between the main investigator and the co-authors (132).

Confirmability refers to a qualitative researcher’s awareness of being influenced by presuppositions – e.g., culture experiences, hypotheses, and theories – and realising that therefore, he or she cannot be considered an objective observer (134). The co-authors were involved as observers during the focus group interviews and in the analysis of the data in both studies. Therefore, they were able to challenge the presuppositions and unconscious assumptions of the main investigator.
7.3.4 Systematic review (paper III)

In this study, meta-ethnography, as described by Noblit and Hare (102), was used to synthesise the results of the qualitative research. Meta-ethnography has been demonstrated to be a useful approach (135), and it has been used in several studies (2, 28, 135, 136). However, many aspects of the phases of the synthesising process remain ill-defined (137), resulting in practical challenges (137) and different interpretations of the approach (2, 28, 135).

One of our challenges was the ability to locate relevant studies. Our research strategy was not able to detect all of the potentially eligible studies because two of the included studies (138, 139) were identified through other sources. In addition, we may have been unable to identify potentially eligible qualitative studies that were published after May 2010 (2).

Another challenge was inclusion decisions. Although we attempted to include studies that focussed on similar populations, different CFS case definitions were used in each study, which may have contributed to differences in the findings of the included studies and may have influenced the overall synthesis. Additionally, many of the studies may be biased because the vast majority of the participants were recruited from healthcare clinics, patient organisations and support groups, and almost all of the studies were conducted in Western countries. The population evaluated in this review may therefore not be representative of all individuals with CFS.

As mentioned in § 7.3.2, the assessment of quality in qualitative research has been subject to debate (100, 128-130). We decided to perform a quality assessment to allow us to exclude studies with very low methodological quality to avoid distorting the overall synthesis (130).

7.3.5 Randomised controlled trial (paper IV)

The quality of quantitative research is usually judged in terms of reliability and validity.

Reliability refers the degree of agreement between two measurement methods or situations, i.e., the reproducibility of the results (140). In our study, reliable and valid outcome measures were applied. Reliability was enhanced by the main investigator having the primary responsibility for conducting all of the analyses in study IV and by regular communication between the main investigator and the co-authors. In addition, a manual for the programme leaders was developed to ensure that each group intervention was delivered using the same approach. Moreover, the programme leaders participated in a three-day seminar focusing on the ability to impart knowledge about CFS, the content of the programme, and the handling of group-processes. This seminar provided the leaders with knowledge and skills needed to
conduct the programme in similar ways.

*Validity* is related to whether you test measures what you desire to measure. Three types of validity may be distinguished: concept validity, internal validity and external validity (140).

*Concept validity* is associated with the concept of being studied and measures the extent to which the problem being studied is operationalised by the use of suitable and adequate variables and tests (140). In the randomised controlled trial (paper IV), the studied concept has been described, and reliable and valid outcome measures were applied. However, the self-efficacy scale (92) had to be translated from Dutch into Norwegian, whereas the Illness Cognition Questionnaire (ICQ) had to be translated from English into Norwegian. This process included an initial translation into Norwegian, a translation back to the original language, and approval using a consensus meeting with some of the translators, as well as testing of the prefinal version (141). Psychometric testing of the final version, as recommended by Beaton and colleagues (141), could not be conducted because of time constraints. This lack of psychometric testing may be a limitation of the present study.

Concerning physical functioning, it is possible that the use of more adaptive coping strategies does not improve physical functioning (105) or lead to improvements over a longer period than was observed in our study. Although the physical functioning subscale of the SF-36 has been used in several other studies (33, 42), physical functioning may have been a less appropriate variable for measuring coping within one year after participation in a self-management programme.

In contrast with our study, other studies have shown that self-management programmes based on self-efficacy may lead to improvements in health status (56, 57, 67, 69). In those studies, the self-rated health item from the Medical Outcomes Studies (142) was used, whereas we used the physical composite summary and mental component summary of the SF-36. The participants in the CFS Empowerment Project (70), which included a self-management programme, also reported improved quality of life (23, 24). In those studies, the Quality of Life Index was used (143). The Medical Outcomes Studies or the Quality of Life Index might have been more suitable instruments for measuring health status in our study.

*Internal validity* concerns the conclusions drawn from the study and is associated with valid inferences regarding the studied population (140). Randomised controlled trials, such as those used in our study, are supposed to have a high internal validity because randomisation and blinding avoid an association between treatment and external factors that may influence the results of a study (confounding) (144). However, bias may occur in relation to the sample
selection bias, information bias and statistical confounding (140).

Concerning the sample selection, the participants were recruited from a variety of sources in different areas in southeast Norway and were included using broad inclusion criteria to obtain a population that was representative of all individuals older than 18 years with mild to moderate CFS in Norway. In addition, our study had a low dropout rate (13.9%), compared with other studies (42). At the one-year follow-up, there were still 59 participants in each group (3 more than calculated in our power analysis), which reduced our risk of making type II errors (140, 144). A significant difference between the intervention and control group was determined for gender. However, analyses of the entire sample showed no significant differences between men and women with respect to any of the outcome measures.

Information bias occurs when the participants in a study report incorrectly or when the registered information is flawed in other ways (140). To reduce the information bias, only one person registered the data, and participants were contacted if any of their data were missing. At the final count, only 0.003% of the possible values were missing. The missing data were replaced using an imputation procedure in SPPS to reduce the information bias.

Concerning statistical confounding, we applied statistical tests that were presented with effect estimates, confidence intervals and p-values (140). The low dropout rate allowed the use of ‘per-protocol’ analyses for within-group comparisons, in which only participants with complete registrations for an outcome measure are included in the statistical analyses of the respective outcome measure.

External validity concerns generalisation and depends on design, population and statistic models (140). In general, randomised controlled studies have a low external validity, although a large study sample and the use of broad inclusion criteria – as in our study – may enhance generalisation (144). Nevertheless, we have strived to present an explicit conclusion to allow the reader to assess the external validity (140).

7.3.6 Challenges in using RCTs to evaluate complex interventions

The outcome measures selected for use in study IV were based on the results of study II, the different theories and concepts upon which our intervention was based, and other studies measuring the effectiveness of interventions based on these theories and concepts, as recommended by the framework for developing and evaluating complex interventions (74). However, discrepancies between the findings in study II and IV raise questions about the extent to which the quantitative methods that were used to evaluate the programme were able to detect the potential effects of the intervention. Wade (145) states that complexity in
healthcare encompasses the existence of multiple factors that influence the variables of the
outcome of interest and the non-linear relationships between the factor and the variable of
interest. This leads to great uncertainty about the causal relationships between individual
items and major unpredictability regarding the effect that may arise following the alteration of
one or more factors. Thus, additional research is needed to establish whether measurements of
the case complexity are useful and if so, in what way it is best achieved (145).

Both the initial and updated framework mention that a process evaluation - using
qualitative studies – may often provide valuable insight into why an intervention fails
unexpectedly or has unanticipated consequences, or why a successful intervention works and
how it can be optimised (74, 76). Initially, a qualitative evaluation after the intervention was
not planned, but we considered the addition of a qualitative evaluation while conducting study
IV. Unfortunately, we were unable to find a master’s student or other researcher who was
interested in conducting this evaluation that could have created supplementary knowledge of
the participants’ experiences with the programme. However, this evaluation may be
performed in a follow-up study.
Summary, conclusions and future suggestions

In conclusion, the evaluation of the developed self-management programme for people with CFS suggests that the programme had a stabilising effect on fatigue severity and self-efficacy at the six-month follow-up. The low dropout rate and the high number of sessions attended may indicate that this programme is an acceptable and helpful intervention. Further evaluations of the effectiveness of group-based self-management programmes for people with CFS are warranted, particularly to enhance coping with the illness.

There are challenges in designing studies to reveal effects of complex interventions. Therefore, a process evaluation and longer-term qualitative and quantitative follow-up studies are needed before firm conclusions can be drawn regarding the effectiveness of our programme. In addition, research is needed to evaluate the cost-effectiveness of the programme (44, 74). Furthermore, measures of complexity in healthcare are still poorly developed, which presents an on-going challenge (145).

Clinical implications

The developed and evaluated self-management programme

- Appears to have a stabilising effect on fatigue severity and self-efficacy at the six-month follow-up.
- Appears to be an acceptable and helpful intervention.
- May be more effective if individual consultations are included.
- May be more effective over time when supplemented with guidance or follow-up after participation to maintain the use of strategies for better coping with CFS.
- May be offered in a primary healthcare setting if follow-up studies show that the programme is an effective intervention
- Further development of the programme may be necessary prior to implementation of the programme in a primary healthcare setting.
- May be led by a juxtaposed occupational therapist and peer counsellor after they have participated in the training programming and if they use the detailed manual.
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Papers I - IV
From chaos and insecurity to understanding and coping: experienced benefits of a group-based education programme for people with chronic fatigue syndrome

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The aim of this study was to elicit participants’ experiences with a multidisciplinary patient education programme and their views regarding the usefulness of the programme. Focus group interviews were conducted with 10 participants immediately and nine months following participation in the programme and analysed using thematic analysis. Initially, the participants experienced confusion and insecurity regarding coping with the illness. Participation in the programme appeared to promote understanding, acceptance and coping through gaining greater knowledge, learning coping strategies and exchanging experiences, as well as receiving understanding and acceptance. Thus, the programme was experienced to be a beneficial intervention. However, the participants expressed a need for more guidance or follow-up to maintain the practice of coping strategies for better coping with their illness.

Keywords: chronic fatigue syndrome; patient education; positive emotions; coping; qualitative research

Introduction

Chronic fatigue syndrome (CFS), also known as myalgic encephalopathy (ME), is an illness associated with physical and cognitive disability. Living with the illness may lead to reduced quality of life (Anderson & Ferrans, 1997), identity crisis (Edwards, Thompson, & Blair, 2007) and social isolation (Drachler et al., 2009; Edwards et al., 2007). CFS is characterised by unexplained chronic fatigue and four or more of the following symptoms lasting at least six months: significant difficulties in short-term memory and concentration, sore throat, swollen lymph nodes, muscle pain, multi-joint pain, new-type headaches, unrefreshing sleep and post-exertional malaise for more than 24 hours after activity (Fukuda et al., 1994). There are no specific tests for diagnosing CFS. Thus, the diagnosis is based on the description of symptoms, the functional level of the patient and by ruling out alternative diagnoses.

CFS has been described in all age groups, social classes and ethnic groups, albeit with a much higher prevalence among women (Jason et al., 1999). The estimated prevalence...
rate of CFS is 0.2–0.4% (CFS/ME Working Group, 2002). Currently, there is no cure or widely accepted treatment for CFS. Although some patients recover, for most of them CFS remains a chronic condition (Brown, Bell, Jason, Christos, & Bell, 2012; Jason, Porter, Hunnell, Rademaker, & Richman, 2011), meaning that these patients must learn how to cope with the illness. Studies have shown that patients have to learn to cope with their illness on their own, mainly by trial and error over time (Edwards et al., 2007; Whitehead, 2006). Given the negative impact of the illness and the lack of curative treatments, it is important to develop effective non-pharmacological interventions that promote better coping abilities.

A few studies have explored group interventions using various approaches. Some programmes offered patient education (Bülow & Hydén, 2003; Carlsen, 2003; Söderberg & Evengård, 2001; Taylor, 2004) or cognitive behavioural therapy (CBT) (Bazelmans et al., 2005; Lopez et al., 2011; Saxy & Hansen, 2005). Other programmes combined CBT with graded exercise therapy (GET) (Belgamwar et al., 2009; Pardaens, Haagdorens, Van Wambeke, Van den Broeck, & Van Houdenhove, 2006) or with GET and relaxation (Kempke et al., 2010). One programme focused on pacing and GET (McDermott et al., 2004). Almost all of these studies reported positive outcomes; however, because they used different types of outcome measures and reported varying drop-out rates, they are difficult to compare. In addition, data obtained from questionnaires provide minimal insight into subjective therapy experiences. Only a few qualitative studies concerning group interventions for people with CFS have been published. Two studies found that the most valuable aspect of their group interventions was the exchange of experiences among participants (Bülow & Hydén, 2003; Söderberg & Evengård, 2001).

At our hospital in South-East Norway, a patient education programme for people with CFS has been offered since 2004. The purpose of the programme is to provide participants with information regarding the illness to promote better coping with the illness, but we are unsure about the usefulness of the programme. The aim of this study was therefore to elicit participants’ experiences with the programme and their views regarding the usefulness of the programme immediately and nine months following participation in the programme.

Methods

Procedure

A qualitative study was conducted using focus groups, given that group dynamics between participants in a focus group may generate a larger and richer range of ideas than individual interviews (Goodman & Evans, 2006; Wibeck, 2000). A major limitation of the method is the risk of individual participants dominating discussion and excluding the contributions of others (Goodman & Evans, 2006; Kitzinger, 1995). Therefore, an important role of the moderator is to encourage discussion and participation and refrain from giving preference to speakers whose views are perceived as the most interesting (Goodman & Evans, 2006).

The focus group interviews were conducted immediately and nine months following participation in the programme. We chose to conduct focus group interviews at these two time points to gain insight into changes in the experienced usefulness of the programme over time. The semi-structured interview guide covered topics such as perceived helpfulness of the different sessions, perceived benefits from the whole programme, ideas regarding the illness, psychological coping processes and applied coping strategies. The interviews lasted 95–110 minutes, and were audio-recorded and transcribed verbatim.
The first author acted as the moderator, and the third or second author was an observer during the first and second interview, respectively.

**Subjects**

Participants in the CFS patient education programme that occurred from February to April 2010 were invited to participate in the study. Information about the study was disseminated through oral and written communication. Nineteen of the 33 participants agreed to participate. Six of these individuals were excluded because their medical record revealed that their diagnosis did not comply with the Canadian diagnostic criteria (Carruthers et al., 2003) and/or the Centers for Disease Control and Prevention criteria (Fukuda et al., 1994) for CFS. Three participants withdrew before the first interview. The final sample comprised eight women and two men. See Table 1 for information on demographics and functioning. The sample represented variations on gender, illness duration and social background. Only one of the participants was working. Six of 10 had higher education, and six were married or lived with a partner.

**Intervention**

The patient education programme was developed, conducted and adjusted several times by health care workers in co-operation with two peer counsellors (i.e. experienced CFS patients), prior to our study. The programme that was evaluated in our study intended to promote coping by providing the participants with information illuminating different aspects of the illness and included the following topics: coping skills, current medical approaches, personal relationships, pacing and energy conservation, physical exercise and relaxation, nutritional approaches and economic self-sufficiency. The programme

<table>
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<tr>
<th>Participant</th>
<th>Age</th>
<th>Illness duration in years</th>
<th>SF-36, subscale physical functioning</th>
<th>FSS</th>
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<td>6.6</td>
<td>42.5</td>
<td>58.5</td>
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<td>Range</td>
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<td>49–63</td>
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*Medical Outcomes Study Short-Form General Health Survey (SF-36), subscale physical functioning (Ware & Sherbourne, 1992), with a 0–100 scale, where 100 is best.

Fatigue Severity Scale (Krupp, LaRocca, Muir-Nash, & Steinberg, 1989), with a 9–63 scale, where 63 is the maximum fatigue score.

Hospital Anxiety and Depression Scale (HADS), with a 0–21 scale for anxiety and depression (Zigmond & Snaith, 1983), where 21 is the maximum score. Cut-off point of 11 is used to identify anxiety and depression (Zigmond & Snaith, 1983).
lecturers were experienced health care workers (physician, occupational therapist, physiotherapist, psychiatric nurse, dietician and social worker) with competence in CFS. Each was responsible for teaching a topic during one of the programme meetings. A peer counsellor shared her own experiences with the illness and useful coping skills during one of the meetings and acted as a positive role model during the others meetings. During the last meeting, the remaining questions from the participants were answered by the lecturers and the peer counsellor. Some of the lecturers focused mainly on information-giving, whereas others combined lectures with group discussions. The participants met eight times over a nine-week period. Each meeting lasted two hours, including a 30-minute break, with the option to engage in relaxation exercises.

**Data analysis**

Thematic analysis followed Richards and Morse’s (2012) description and involved a series of steps. First, the interviews were read thoroughly to form a general impression of their contents. Second, topic coding was used to identify and label sentences and paragraphs on a topic. Third, topics that appeared to pertain to a similar phenomenon were compared and related to each other. Finally, abstraction was achieved by gathering topics in categories. The second, third and last steps were applied as an ongoing flexible process and did not follow a linear sequence. The analysis was primarily conducted by the first author, whereas the second author was involved in the last step. The emerging categories that related to coping with the illness were ‘chaos and insecurity’, ‘understanding’, ‘acceptance’ and ‘coping’. Categories comprising mediating factors were ‘knowledge about the illness’, ‘knowledge about coping strategies’, ‘exchange of experiences with fellow participants’ and ‘receiving understanding and acceptance’.

**Ethical issues**

The study was approved by the Regional Committee for Medical Research Ethics (Health Region South-East) and the Privacy Ombudsman for Research. Written informed consent was obtained from all participants who were also informed of their right to withdraw from the study. To protect the anonymity of the participants, information that may lead to personal identification has been removed.

**Results**

The results of the analysis elucidated the process of how the participants experienced coping with their illness before and after participation in the programme, and how the programme facilitated this coping process in different ways (see Figure 1). The phase before commencing the programme was characterised by experiences of chaos and insecurity. In the period after participation in the programme, the participants experienced better understanding and progression in acceptance and coping processes.

**Experiences of chaos and insecurity**

Before commencing the programme, the participants experienced problems with understanding that they were ill: ‘I was in denial. I am fit, especially on good days, and if I wasn’t fit I felt I was very lazy, good-for-nothing, and I wanted to be fit’.
Several participants lacked information regarding how CFS was diagnosed, and some were therefore not convinced that they had CFS. In addition, they were unsure regarding the prognosis of their illness. Some had the impression that it was possible to achieve a full recovery after some time, whereas others considered CFS to be a chronic condition. Hope of improvement over time, without the expectation of full recovery, was also expressed.

All participants expressed some degree of confusion regarding how to cope with the illness. Some had not received any information or help after being diagnosed, whereas others had received advice that made them feel worse: ‘It worked for me before that if I used my body, I gained energy. Physicians told me the same thing, that you have to activate yourself, but that worked out very badly’.

Those who had received individual guidance experienced difficulty with translating new knowledge into practice because of insufficient follow-up by health care professionals. Most of the participants felt as though they had been left alone with the illness: ‘Before the course I felt very alone, I had no one to talk to’.

Experiences of understanding, acceptance and coping
Immediately following the programme, participants expressed more positive emotions, such as feeling more tranquil and confident than before. They reported better understanding and being in an acceptance process. Especially after nine months, the participants experienced better coping with their illness.

Better understanding
Soon after the programme, participants realised that they actually were ill, and some expressed greater confidence regarding their diagnosis and also greater awareness that
their symptoms were related to CFS: ‘I think I have a lot more understanding of the illness and all these strange symptoms I had that scared me to death’.

Some participants felt more able to assess information about the illness more critically: ‘The course helped me to create my own room, to create walls that enabled me to extract relevant information, because I think if you want to try everything, you eventually will become insane’.

In addition, the participants had become more critical towards all types of ‘miracle cures’: ‘I think that regarding alternative treatment, you try different things, keep looking for a solution. I have become more relaxed after the course, and have stopped trying new things all the time’.

Acceptance
Immediately following the programme, the participants also described a change in their understanding of the illness trajectory. Some of them had expected that participation in the programme would cure them but then realised that they actually had to focus on acceptance and coping with the illness: ‘I expected to become fit, but after the course I realised that I had to begin to accept. Well, I have actually started the process, so that is positive’. Others experienced increased acceptance of the illness. After nine months, all participants expressed increased acceptance, although at times some still felt that acceptance was equivalent to giving up on the hope of getting better.

Coping
Although some still hoped for recovery, they realised that better coping was a necessary step towards recovery, and that recovery could take a long time. Immediately following the programme, participants felt that they had gained new insights and understandings, and envisioned new ways of coping with the illness. Nine months later, they had begun to use new coping strategies in daily living, although to varying degrees. They experienced better coping with their illness and an increased feeling of control, but did not experience better health.

Most participants believed that they had gained better insight into the relationship between activity level and symptom severity, and had learned that overexertion could lead to exacerbations of their illness. In addition, many participants felt better able to cope with such exacerbations: ‘When I am quite exhausted I have learned that I have to take it very easy, lie down until my body says “now it’s better, now it’s enough”’. Better coping was also experienced with regard to accompanying anxiety and depression:

All this restlessness and anxiety and things like that come up in between anyway, but then you know that it probably will pass when you use techniques or just take it very easy. I have become much more able to take it very easy.

Some participants had become more conscious and gained a greater understanding of associations between performed activities and fatigue. They had begun to analyse what may have exacerbated their illness to prevent or limit the extent of future deteriorations:

It still is detective-work. When I get fatigued, I easily start analysing what I have done. I have never analysed in such a conscious way before. I have found out that it varies. Some activities wear me out, but only for a short period of time and it doesn’t get worse, while other activities wear me out even in small amounts.
Resting more often than they were accustomed to, which resulted in a better balance between activity and rest, was experienced to decrease deterioration in their condition or prevent deterioration. Some participants took breaks when they became tired, whereas others paced their activities according to time schedules. In addition, participants felt more comfortable with resting than before. Different relaxation techniques were applied to improve the quality of their resting breaks.

The participants also gained better insight into the amount of energy that different activities require and thus felt more able to prioritise the use of their energy and to make deliberate choices regarding occupational participation than before. This occasionally included saying ‘no’. One participant noted: ‘When I’m driving my car for instance, I notice that I have to pull myself together, and I have become more aware that I can lose focus when driving longer distances. This is such an aha-experience for me’.

Practising energy conservation was experienced as beneficial, and participants presented several examples of what they had done to make the performance of daily tasks less exhausting:

I only do my shopping in shops where I can park right outside and carry my groceries straight into my car. I try to work at waist-height and always place grocery-bags on a chair. I sit on a bar stool in front of the stove when making pancakes.

In addition, some participants had begun to use assistive devices in daily living to conserve energy, such as shower stools, work chairs and wheelchairs.

A better understanding of the importance of eating healthy food was reported, and several subjects had changed their diets. Some had experienced benefits from spreading their meals over the day to assure a more frequent energy supply, whereas others had begun to drink more water during the day. In addition, the consumption of foods with low carbohydrate content was found to reduce blood sugar fluctuation. However, some individuals felt unable to change their diets because they lacked the appetite or the energy to do so.

Some of the participants felt more confident talking about the illness with others than before, whereas others still preferred to avoid the subject. In addition, participants had started to use new strategies when dealing with other people’s misunderstandings and negative attitudes towards CFS. For example, participants related that other people took their illness more seriously when the participants referred to receiving treatment for CFS at a hospital. Some subjects had increased their acceptance of other people’s insufficient understanding of, or lack of interest in, CFS:

I know that you should explain and not care about what others mean. I have been thinking a lot about this, that this is my illness and it is just me who knows how I feel, and I have learned that others actually are unable to understand.

Immediately following the programme, several participants felt a need for more guidance or follow-up to maintain the practice of coping strategies for better coping. After nine months, several participants had tried to obtain help, but had found that it was difficult to find primary health care professionals with competence in CFS.

**Mediating factors**

Participants experienced that the programme promoted understanding, acceptance and coping through gaining greater knowledge about the illness and coping strategies,
exchange of experiences with fellow participants and receiving understanding and acceptance.

Knowledge about the illness
Learning how the illness was diagnosed and learning about the symptoms and possible causes increased understanding and confidence regarding the individual’s own condition: ‘Through the course I have become more and more sure of ME probably being the right diagnosis, because I have seen and heard others, and got a full presentation of the criteria for the diagnosis’.

The participants also learned about the prognosis and that it could take a long time to improve. In addition, it was considered helpful to learn that deterioration may occur even when doing everything ‘right’.

Knowledge about coping strategies
Participants found it especially helpful to learn about pacing and energy conservation: ‘I had absolutely no comprehension of how demanding different activities can be; that you use much more energy bending forward or working aloft compared to working at waist-height. Now I can take care of things like that’. Other topics considered beneficial were the practice of relaxation exercises and learning how to deal with difficult feelings. In addition, the participants appreciated receiving information regarding economic and public support systems and nutrition.

Exchange of experiences with fellow participants
All participants found it helpful to exchange coping experiences and share beneficial coping strategies. For some participants, this was the most valuable part of the programme: ‘It was very good to meet others that are in the same situation. You learn a lot and become more conscious through others. And they have tips: “I have tried this, I have tried that and that”’.

Receiving understanding and acceptance
It was an overall positive experience for the participants to receive understanding and acceptance from fellow participants that were experiencing the same type of symptoms and problems: ‘And it was very great to be together with people that understand the condition, finally being at a place where it feels normal’.

The participants felt that mutual understanding made it safe to discuss issues they had not been able to discuss elsewhere:

It was so good just to open up completely like you are unable to do at home. I feel in a way that when we had finished talking and I went home, I had left it all behind, and I could live a bit more at home again.

The presence of a peer counsellor increased the feeling of safety and fellowship, and she was valued as an important role model. In addition, participants appreciated meeting health care professionals with knowledge of CFS: ‘It made me feel safe to come to a group and health care professionals that understand my condition and respect it’.
Discussion and conclusion

The present study showed that participation in the programme appeared to promote understanding, acceptance and coping through gaining greater knowledge, learning coping strategies, exchanging experiences and receiving understanding and acceptance.

Before participation in the programme, the participants experienced confusion and insecurity regarding their condition, partially because of the lack of information and emotional support from their GPs and other health care professionals after being diagnosed with CFS. This phenomenon has also been reported in other studies (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012; Edwards et al., 2007; Horton et al., 2010). Similar to findings in other studies (Dumit, 2006; Edwards et al., 2007), participants felt as though they had been left alone with the illness and lacked strategies that enabled them to cope with their condition. This result may be explained by a possible downward spiral between negative emotions and maladaptive coping strategies, as found by Burns et al. (2008).

Immediately following the programme, participants expressed feeling more tranquil and confident than before. They had gained new insights and understandings and experienced increased acceptance that made it possible to see and learn new methods of coping with the illness. After nine months, the participants had begun to use new coping strategies in their daily living, although to varying degrees. These findings were consistent with studies showing that positive emotions may broaden thought-action repertoires (Fredrickson & Branigan, 2005) and may be associated with increased coping (Burns et al., 2008). Moreover, several other studies concerning CFS have found that understanding and accepting the illness seems to facilitate coping (Edwards et al., 2007; Gray & Fossey, 2003). Acceptance may promote the downgrading of expectations and the search for new sources of fulfilment (Drachler et al., 2009; Edwards et al., 2007; Gray & Fossey, 2003). Acceptance seemed to be a fundamental component in shaping the participants’ adjustments to living with CFS. The fact that the participants in our study described varying degrees of using new coping strategies after nine months may indicate that the programme needs to focus more on progress in acceptance processes to facilitate the use of adequate coping strategies, and indicates a need for follow-up after participation in the programme.

Participants reported that the programme had given them useful tools to deal with other people’s disbelief and misunderstandings. Not being understood by others is an often reported issue that may lead to withdrawal from social life and a loss of friendships (Åsbring & Närvänen, 2002; Clarke & James, 2003; Drachler et al., 2009; Edwards et al., 2007), therefore, it may be beneficial to add a meeting for relatives to the programme to improve their knowledge about the illness.

Pacing of activities was emphasised by the participants as an important coping strategy. The usefulness of pacing is disputed among scientists, but several qualitative studies support the findings in our study (Anderson et al., 2012; Clarke & James, 2003; Drachler et al., 2009; Edwards et al., 2007; Whitehead, 2006). Our findings were also consistent with the results of a survey sponsored by The ME Association (2010).

Other qualitative studies (Edwards et al., 2007; Gray & Fossey, 2003; Whitehead, 2006) have shown changes over time, similar to those in our study. The changes observed in the present study, however, appeared to be related to participation in the patient education programme through gaining greater knowledge, learning coping strategies, exchange of experiences with fellow participants and receiving understanding and acceptance.
Some studies have revealed that sufferers desire fellowship with other CFS patients for support, sharing of experiences (Åsbring & Närvänen, 2002; Carlsen, 2003; Dumit, 2006) and sharing of information (Carlsen, 2003; Dumit, 2006), e.g. in support groups and online. Åsbring and Närvänen (2002) observed that this sharing is not necessarily a positive experience for all patients. One reason that sharing was a positive experience in the present study may be that the meetings were conducted by health care professionals who focused on constructive solutions more than negative experiences.

Studies have shown that patients have to learn to cope with their illness on their own (Edwards et al., 2007; Whitehead, 2006). Therefore, patient education programmes where coping strategies can be presented and exchanged among participants may be useful to provide organised information. In some studies, participants felt that if they had received advice on how to manage their illness during the early stages, they would not have become as distressed (Edwards et al., 2007; Pheby & Saffron, 2009), which may indicate a need for offering patient education programmes shortly after diagnosis of CFS.

One important limitation of our study was that the first author had two individual consultations with some of the participants prior to the patient education programme and was involved in the programme as a teacher during one of the eight meetings, which may have influenced the extent to which the participants felt free to express their negative experiences. Qualitative research requires an accurate description of the participants’ experiences. Therefore, the participants were informed that their anonymity was assured and that participation in the study would not have consequences for future treatment at our hospital. In addition, the relationship between the researcher and the participants can become pseudo-therapeutic and the researcher may have difficulties in separating own experiences from those of the participants; this may result in subjectivity (Jootun et al., 2009). Therefore, the first author had to make an effort to focus solely on the participants’ experiences to produce theoretical understanding and sensitivity (Jootun et al., 2009). In this way, she was able to construct meaning that would not be apparent to an external moderator. Another limitation was that the participants were recruited from individuals who were taking part in the patient education programme at the time; less than half of the programme participants took part in this study. Thus, the study population may not be representative of the total population that may usually participate in our patient education programme. However, the baseline scores on the questionnaires suggested that the participants were comparable to participants in similar studies (Jason et al., 2011; Jason & Brown, 2013; White et al., 2011).

People with CFS have a need for education to enable them to understand, accept and cope with their illness. Only a few qualitative studies have been published providing insight into subjective experiences with group interventions. The patient education programme in this study was experienced to be beneficial. However, the participants expressed a need for more guidance or follow-up to maintain the practice of coping strategies for better coping with their illness. More research is needed to ensure that these findings are representative of the experiences of other people with CFS. In addition, the effects of the programme should be evaluated in a larger quantitative controlled study.

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Development of a group-based self-management programme for individuals with chronic fatigue syndrome: a pilot study

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Abstract
Objective: The aim of the study was to develop a group-based self-management programme for individuals with chronic fatigue syndrome (CFS) by using the participants’ experiences with the initial version of the programme, which intends to promote coping with the illness in a primary healthcare setting. Methods: An initial programme was developed, based on self-efficacy theory and the concepts of client-centred practice and empowerment. Subsequently, the programme was tested and further developed by drawing on the participants’ experiences with the programme. Focus-group interviews were applied. The interviews were analysed using thematic analysis. Results: The initial programme was found to be feasible, although several modifications regarding the content and practical organization of the programme were proposed. Conclusion: In line with the participants’ experiences, the final self-management programme was developed, which includes short presentations of eight topics, exchange of experiences among participants, goal-setting, construction of action plans, and relaxation exercises, in addition to a meeting for relatives. The programme will be provided in eight biweekly sessions and be led by juxtaposed peer counsellors and occupational therapists. The effects of the final programme will be evaluated in a randomized controlled trial.

Key words: self-efficacy, qualitative research, patient education, empowerment, coping, complex interventions, client-centred practice

Introduction
Chronic fatigue syndrome (CFS), also known as myalgic encephalopathy (ME), is a disabling condition that affects different aspects of everyday life, including social and vocational participation (1,2). Living with this illness may lead to reduced quality of life (3), identity crisis (1,4), and social isolation (1–4). CFS is characterized by unexplained chronic fatigue and four or more of the following symptoms for at least six months: impaired short-term memory and concentration, sore throat, swollen lymph nodes, headaches, muscle pain, multi-joint pain, and post-exertional malaise for more than 24 hours after activity (5). The estimated prevalence of CFS within the general population is 0.2–0.4% (6), with a higher prevalence among women (7). There is currently no known cure for this condition. Some patients fully recover, whereas others progressively worsen. Most patients function at a stable but reduced level most of the time (8).

Several studies have shown that patients receive insufficient information, help, and support from healthcare professionals after diagnosis (1,9). Drachler and colleagues (9) found that CFS patients have a need (i) to make sense of symptoms; (ii) for...
information on the illness; (iii) to develop strategies to manage impairments and activity limitations; and (iv) to develop strategies to maintain and regain social participation. Given the negative impact of this illness and the lack of curative treatments, effective interventions need to be developed to promote better skills for coping with the illness. The potential role of patient education programmes will likely become increasingly important.

Only a few studies have been published concerning patient education for individuals with CFS. Taylor and Jason conducted the CFS Empowerment Project and found that this consumer-driven programme had a positive impact on symptom severity (10), quality of life (10,11), and resource acquisition (12). Two qualitative studies have shown that the most valuable aspect of group interventions for CFS patients is the exchange of experiences among participants (13,14).

In Norway, the organization of healthcare is guided by the Coordination Reform, which is based on equal access to high-quality healthcare services, regardless of one’s place of residence (15). This reform emphasizes “coping” and mentions establishing community-based patient education programmes as one of several potential actions. However, several studies have indicated that patient education alone is not sufficient to improve clinical outcomes (16,17). Rather, programmes teaching self-management skills are more effective in this regard (18). A self-management programme allows patients to identify their problems and provides techniques to help patients make decisions, take appropriate actions, and alter these actions as they encounter changes in their circumstances or disease course. Importantly, self-management education complements, rather than replaces, traditional patient education (18).

None of the known patient education or self-management programmes for individuals with CFS can be implemented in primary healthcare in Norway without modifications, because of the specific organization of healthcare, and limited access to healthcare professions with competence in CFS in primary healthcare. The aim of this pilot study was therefore to develop a group-based self-management programme for individuals with CFS by using the participants’ experiences with the initial programme, which intends to promote coping with the illness in a primary healthcare setting in Norway. The effects of the final programme will be evaluated in a randomized controlled trial.

Material and methods

A self-management programme is a complex intervention, because it contains several interacting components (19,20). It can be difficult to determine the effectiveness of complex interventions, especially when an adequate description of the development and content of the intervention is missing (21,22). Therefore, the Medical Research Council in the United Kingdom developed a framework for developing and evaluating complex interventions to improve health (21), which was updated and extended in 2008 (20), to help researchers to recognize and adopt appropriate methods. The key elements of the framework are development, piloting, evaluation, and implementation. The stages in the process may be applied as an ongoing flexible process and do not follow a linear or cyclical sequence (20). We were inspired by this framework when developing and piloting the initial programme in this study. These stages will be described below.

Because the framework recommends thinking about implementation at an early stage (20), an occupational therapist in primary healthcare (LLH) was involved in the whole process. In addition, users should be involved at all stages of the development process as this is likely to result in a better and more relevant intervention (20). Therefore, a representative of the Norwegian ME Association (HHA) was involved in the whole process and participants were invited to share their experiences after taking part in the programme.

The study was approved by the Regional Committee for Medical Research Ethics (Health Region North) and the Privacy Ombudsman for Research at Oslo University Hospital. Written informed consent was obtained from all participants. To protect the anonymity of the participants, information that could lead to personal identification has been removed.

Development of the initial programme

An essential early task is to develop a theoretical understanding of the likely process of change, by drawing on existing evidence and theory (20).

The initial self-management programme is based on the concepts of client-centred practice and empowerment, like the earlier mentioned CFS Empowerment Project (10-12). In client-centred practice there is a strong emphasis on a collaborative partnership, respect for the client, facilitating choice, and involving the client in determining occupational goals that emerge from his or her choices (23). In addition, it stresses the need to equalize power in the clinician-therapist relationship (23). Client-centred practice has been shown to improve levels of satisfaction with services (24) and the ability of patients to recall their goals (25). In addition, client-centred approaches have been reported to improve symptoms and functional outcomes (25,26). Empowerment refers to providing individuals with the knowledge...
and resources to take control of their lives (27), including the promotion of self-reliance and the use of peer networks for solving problems (28).

In addition, the initial programme is based on the self-efficacy theory, which states that “perceived self-efficacy refers to beliefs in one’s capabilities to organize and execute the courses of actions required to produce given attainments” (29). It has been shown that self-management programmes for individuals with chronic illness based on the self-efficacy theory may lead to improvements in health behaviour, health status, and self-efficacy (30-32). To our knowledge there are no publications addressing interventions based on self-efficacy theory for individuals with CFS.

The practical organization of the programme was influenced by a qualitative study evaluating the patient education programme for CFS patients at Oslo University Hospital (unpublished). The results of this study indicated that eight biweekly sessions of 2.5 hours’ duration was suitable. In addition, participants expressed that it was beneficial to exchange experiences with fellow participants and that they appreciated meeting both peer counsellors and healthcare professionals as group leaders.

Based on the presented theories and evidence, we developed the initial programme (Table I). During the programme, coping capabilities are developed through exchange of knowledge and experiences among participants, modelling of self-management skills, guided mastery practice, and informative feedback (29). The programme covers a number of topics chosen by the participants. At the beginning of the programme, the participants set personal goals for coping and construct action plans to achieve these goals. During each session, action plans are evaluated, and the participants are offered feedback from the other participants in overcoming obstacles they faced.

At the end of each session, the current action plans are adjusted if necessary or replaced with new action plans. Finally, a manual for programme leaders was developed.

The initial programme was tested in the piloting stage, as described below. The experiences of the group leaders and the participants resulted in development of the final programme.

**Piloting of the initial programme**

The piloting stage includes testing interventions for their feasibility, acceptability, and beneficence (20).

The initial programme was conducted in the spring of 2011. Participants were recruited by sending brochures to physicians, other healthcare professionals, and members of CFS patient organizations in a mid-sized town and the surrounding municipalities in south-east Norway. All participants were required to have been diagnosed with CFS by a medical practitioner, to be at least 18 years of age, and considered to be physically capable of attending the scheduled meetings. Two males and 16 females were included in this pilot study. One woman withdrew due to a death in the family, and a second woman was too ill to meet. One participant did not finish the programme because of an illness in her family, and another participant did not believe that the programme met her needs. The median number of sessions attended was six (range 1–8).

Initially, the programme was led by IP, LLH, and HHA. However, by the end of the programme, IP acted primarily as an observer to determine whether the programme could be led by one occupational therapist and one peer, as intended.

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**Table I. Content of the initial programme.**

<table>
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<tr>
<th>Days 1</th>
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<tbody>
<tr>
<td>Information about the programme: content and form</td>
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<tr>
<td>Participants and leaders introduce themselves</td>
</tr>
<tr>
<td>Educational presentation about current medical approaches</td>
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<tr>
<td>Educational presentation about self-management</td>
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<tr>
<td>Selecting individual goals and constructing action plans to achieve these goals</td>
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<tr>
<td>Participants identify topics relevant for better coping with CFS and vote on the seven topics they consider to be most relevant</td>
</tr>
<tr>
<td>Two participants take responsibility for preparing a short presentation or questions for dialogue concerning each topic. They may involve one of the leaders or an external lecturer</td>
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<tr>
<th>Days 2–8</th>
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<tbody>
<tr>
<td>Participants present and evaluate their individual goals and action plans with feedback/problem-solving</td>
</tr>
<tr>
<td>Educational presentation in one of the chosen topics by participants with exchange of individual experiences</td>
</tr>
<tr>
<td>Constructing action plans</td>
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Focus-group interviews. The participants were invited to participate in focus-group interviews to share their experiences after taking part in the initial programme. Focus-group interviews were chosen as group dynamics between the participants may generate a larger and richer range of ideas than individual interviews (33,34).

One male and five females were willing to participate in the focus-group interviews immediately following participation in the programme and four months later. The sample represented variations in gender, illness duration, and social background. The mean age of the participants was 38.7 years (range 28–46) and they were diagnosed with CFS from one week to seven years (mean 2.3 years) ago. Only one of the participants was working. Four had higher education, and five were married or lived with a partner.

The semi-structured interview guide that was used during the first focus-group interview covered themes such as perceived benefits from participation in the programme, aspects of the programme that were experienced to be helpful, aspects that were experienced to be less helpful, and suggestions to improve the programme. The results of the first focus-group interview were respondent validated during a second focus-group interview conducted four months later. Proposed changes in the programme were discussed with the participants, in addition to perceived benefits from participation in the initial programme to gain insight into developments over time.

The focus-group interviews lasted from 30 to 90 minutes. All interviews were recorded and transcribed verbatim. The first author acted as the moderator, and the second author was an observer during the first interview, while the second and third author were observers during the second interview.

Analysis of the focus-group interviews. Thematic analysis of the focus-group interviews followed Richards and Morse’s (35) description and involved a series of steps. First, the interviews were read thoroughly to form a general impression of their contents. Second, topic coding was used to identify and label sentences and paragraphs on a topic or theme. Third, topics that appeared to pertain to a similar phenomenon were compared and related to each other. Finally, abstraction was achieved by gathering topics in categories. The analysis was primarily conducted by the first author, whereas the second and third author were involved in the last two steps. The emerging categories that were related to coping with the illness were “insight and understanding”, “acceptance”, and “practising coping strategies”. Categories comprising mediating factors related to participation in the programme were “knowledge”, “meeting fellow patients”, “changing coping strategies”, and “practical organization”. The last category was “meeting for relatives”. The categories are presented below with illustrative quotations.

Results of the focus-group interviews

The participants’ experiences could be gathered in categories that were used in the development of the final programme. Analysis revealed that the two focus-group interviews focused on the same topics, although topics and categories related to “coping with the illness” showed developments over time. Opinions and proposals concerning “mediating factors related to participation in the programme” and “meeting for relatives” became more explicit, concrete and unanimous during the second focus-group interview. Therefore, only the final opinions and proposals will be presented.

Coping with the illness

Immediately following the programme, participants reported having gained new insights and understandings, being in an acceptance process and practising coping skills. The reported benefits were experienced immediately following the programme, but they were particularly evident after four months.

Insight and understanding. The participants felt that they had gained new insights and understandings through the exchange of knowledge and experiences with fellow participants and the programme leaders. They expressed having increased their awareness that their symptoms were related to CFS. One participant said: “I had a lot of questions, and then I got: ‘Yeah, that’s how it is! Yeah, it’s like that for me too!’”. They also expressed having learned that changes in their diets, avoidance of over-exertion, and energy conservation might improve coping with the illness, but that deteriorations might occur even when practising appropriate coping strategies. An improved understanding of the illness and awareness that other people with CFS are experiencing similar symptoms was reported to promote a feeling of being more confident and relaxed, for instance when meeting other people’s scepticism towards the illness. As one participant said: “What others think is not that important any more. That has calmed me down”.

Acceptance. Participants also expressed an increased acceptance compared with before participation in the programme. One participant said: “Now I understand that I’m ill and just have to make the best of it”. After
four months, most of the participants reported progression in accepting processes, but at times some of them could still struggle with themselves, thinking that they were lazy and had to pull themselves together.

Practising coping strategies. All participants reported better coping by practising coping strategies, such as pacing and energy conservation, resting more often, changing their diets, training core muscles, and implementing sleep routines, especially after four months. One participant said: “I feel that I’m having more consideration for myself than ever before. And I’ve become better at telling others when I’m tired, and say ‘no’ if there’s something that I can’t do”.

Mediating factors related to participation in the programme

Several aspects of the initial programme were experienced as helpful, but the participants also proposed several modifications regarding the content and practical organization of the programme. Categories comprising mediating factors related to participation in the programme were “knowledge”, “meeting fellow patients”, “changing coping strategies”, and “practical organization”.

Knowledge. Receiving further knowledge regarding the topics highlighted through presentations and discussions was reported to contribute to increased understanding and insight regarding the participant’s own condition. All participants felt that the topics illuminated during the programme were relevant, but some felt that some of the topics could have been illuminated more thoroughly. No important topics were perceived to be missing.

Participants reported appreciating being able to choose topics themselves, but they had also been worried about missing important topics. Some had experienced having problems with choosing topics because of cognitive impairments. During the interviews, the participants learned that the topics they had selected were similar to the topics that were part of the CFS patient education programme at our hospital. The participants therefore suggested that the programme could feature fixed topics with the possibility of focusing on the participants’ specific challenges during the discussions. They debated and eventually reached a consensus regarding the eight topics that should be covered: current medical approaches, activity pacing and energy conservation, sleep and relaxation, physical exercise, nutritional approaches, economic self-sufficiency, personal relationships, and available treatments. Most of the participants felt that they did not have the knowledge or energy to properly prepare the topic for which they had taken responsibility; as one participant said: “I think we got a little too much responsibility. I felt a lot of pressure even though we were told it only should be a plain presentation”. In addition, participants were unsure of the validity of the information they shared because of conflicting information regarding the illness that is portrayed in the media. They therefore appreciated that a professional from the CFS/ME centre was involved to provide “quality assurance” for the information that was shared during the sessions. The following proposal was made by one of the participants and supported by the others: “I think the competence at the CFS/ME centre should be used by letting healthcare professionals at the centre make short presentations about different topics that can be presented by one of the local course leaders”. Presentations of approximately 15 minutes in duration were considered suitable because they allowed sufficient time for discussing particular problems and for the exchange of experiences. In addition, they suggested distributing some information before each session for preparation, and handing out the presented information and providing tips regarding where to find additional information at the end of each session.

Meeting fellow patients. All participants found it helpful to meet fellow patients who were experiencing the same type of symptoms and problems for exchange of knowledge and experiences, and receiving understanding and acceptance. The participants felt that mutual understanding made it safe to discuss all kinds of issues. One participant said: “When you come to a group where you can air all kinds of things, it helps in many ways. You get more knowledge from everyone. You don’t feel alone any more and relieve some stress”. Several of the participants had felt as though they had been left alone with the illness. During the programme they established contact with fellow participants and reported having met or phoned fellow participants for help and support after they finished the programme. In addition, they appreciated that they had formed a network and about 10 of them had met once a month after they finished the programme.

The peer counsellor was regarded as a valuable role model. One participant said: “I think it’s important to meet someone who has lived with CFS for many years and can give you good advice, someone who through the years has become more able to cope with the illness in daily life”.

Changing coping strategies. Most of the participants found it helpful to practise new coping skills through
goal-setting and the use of action plans; as one participant explained: “Action plans made me focus on things that I had thought about before. It became easier to change these things when I wrote it down and began to use action plans”. Nevertheless, several participants experienced problems understanding the purpose of goal-setting and using action plans. Some difficulties were also reported in the identification of relevant and realistic action plans and their incorporation into daily life without exceeding their limitations. The participants made several suggestions about how goal-setting and making use of action plans might be introduced in a clearer way. They proposed that goals and action plans should reflect changes in coping with challenges related to daily life to promote compliance. They also suggested that it should be emphasized that action plans can focus on doing something new, refraining from doing something that does not promote coping or doing things in a different way. In addition, the participants recommended illustrating the introduction of goals and action plans with several realistic examples.

Receiving feedback from the other participants in overcoming obstacles they faced was viewed as being valuable: “Often you can’t see all solutions yourself, and then it is very useful to get advice from others instead of saying I can’t manage this”.

Practical organization. The organization of the programme into eight biweekly sessions of 2.5 hours’ duration in the middle of the day was considered appropriate.

Participants reported experiencing a lack of flow in the sequences of the course elements. They therefore suggested scheduling educational presentations with an exchange of personal experiences on the topics before the break and to include problem-solving and the development of action plans after the break.

Participants appreciated the fact that the leaders had expertise in CFS and were meeting the participants’ specific needs respectfully. They also valued that the leaders strove for quiet surroundings with resting facilities, dimmed lights, and a comfortable indoor climate.

In addition, participants valued that the programme was conducted in a primary healthcare setting allowing them to meet fellow patients from their neighbourhood.

Meeting for relatives

The participants did not mention the need for a meeting for relatives themselves, but when asked they did state that their relatives need information to be more able to understand the illness. One participant said: “I know that my relatives try to read everything about CFS, but I think they would have appreciated meeting other relatives. Then I think it’s important to have discussions, not only presentations”. They acknowledged that children need information about CFS too, but said that a meeting for adults should be prioritized.

Discussion

This article describes the development of a group-based self-management programme for individuals with CFS that promotes coping with the illness in a primary healthcare setting. We were inspired by the framework developed by the Medical Research Council in the United Kingdom (20) when developing and piloting the programme. In this process, existing theory and research-based evidence was integrated with clinical expertise and the preferences, concerns, and experiences of participants (20).

After participation in the initial programme, the participants reported improvements in their understanding of CFS and their abilities to accept and cope with their illness. These results may have been promoted by guided mastery practice and informative feedback – in terms of using action plans – as Bodenheim and colleagues (18) have reported that programmes with self-management action plans are more effective in improving outcomes than those without action plans. Consequently, actions plans may be included in the final programme.

Initially, the participants experienced problems understanding the purpose of goal-setting and the making of action plans, which may be partially explained by difficulties in short-term memory and concentration that characterize the illness (5). Given the cognitive challenges facing CFS patients, it is critical that the information provided to these individuals be conveyed in a clear manner, repeated several times, and illustrated with concrete examples to facilitate patient comprehension and knowledge retention. The manual for future programme leaders needs to be adapted accordingly.

The reported improvement in coping with the illness may also have been promoted by exchange of knowledge, as several studies have found an association between the acquisition of knowledge and improved understanding and acceptance (36). This may indicate a need to include exchange of knowledge in the final programme.

Pacing of activities was emphasized by the participants as an important coping strategy. The usefulness of pacing is disputed among scientists, but several studies support the finding in our study (1,2,10). Pacing may therefore be included as a topic in the final programme.
The participants initially appreciated being empowered by choosing the topics covered during the programme. However, when they learned that the topics they had chosen were similar to those covered in the CFS patient education programme at Oslo University Hospital, they reported feeling that these topics were sufficient to fulfil their needs and therefore concluded that choosing topics did not need to be included in the final programme. The chosen topics were similar to the topics that were covered in the CFS empowerment project (12) as well. This result may indicate that these topics are those that most concern CFS patients and ought to be included in any self-management programme. However, new knowledge about CFS may change CFS patients’ needs over time.

In this study, the preparation of presentations was reported to be too exhausting. This is not a surprising result as CFS is characterized by chronic fatigue and cognitive impairments. In addition, the participants felt unsure with regard to the extent they could trust the information they had found. Thus, they proposed that healthcare professionals at the CFS/ME centre make short presentations covering fixed topics. Using presentations made by external healthcare professionals may provide “quality assurance” of the imparted information, and at the same time allow enough time for exchange of information and experiences that will meet the participants’ particular needs. The proposed change might strengthen the client-centredness of the intervention, as adaptation of the intervention might meet the participants’ needs in a better way and might enable them to make better informed choices (23). However, this change entails a need for updating the presentations to include new knowledge.

The participants confirmed that modelling of self-management behaviours can be accomplished by having peers teach self-management programmes (37). Several studies have shown that peers, when well trained and provided with a detailed manual, teach as well as healthcare professionals (30,38). It may therefore be beneficial to include peer counsellors as leaders in the final programme.

Many CFS patients experience difficulties with their diagnosis being properly acknowledged and understood by healthcare professionals (1,2). Consequently, the participants appreciated that the programme was co-led by healthcare professionals with competence in CFS. It may, however, be an advantage that occupational therapists are involved as programme leaders as several of the chosen topics contain issues that may be part of occupational therapy interventions, for example activity pacing, energy conservation, physical exercise, and available treatments. Consequently, self-management programmes may preferably be conducted by juxtaposed peer counsellors and occupational therapists.

An occupational therapist in primary healthcare was involved at an early stage to ensure implementation of the intervention (20). In addition, the participants appreciated that the programme was organized in a primary healthcare setting. Thus, it may be possible and acceptable to organize the self-management programme in a primary healthcare setting.

One problem frequently reported by CFS patients is not being understood by their significant others (1,9). As a result, patients can feel lonely, isolated, and abandoned. It is therefore not surprising that the participants supported including an information meeting for relatives within the final programme.

Our study has several limitations. The first three authors were both programme leaders and involved in the focus-group interviews as moderator or observer, which may have influenced the extent to which the participants felt free to express their negative experiences. However, as the first three authors had been involved in the development and piloting of the programme they may have been more able to generate a richer and deeper evaluation compared with an external moderator.

In addition, it is important to consider that data from qualitative research can be understood and interpreted in more than one way. Inevitably, the results presented may be shaped by the characteristics and perspectives of the authors (occupational therapists and peer).

Finally, only a minority of those who participated in the programme were willing to take part in the focus-group interviews. Thus, the responses obtained from the focus-group interviews may not be representative of all participants. However, the information obtained through the focus-group interviews was largely congruent with the experiences of the programme leaders.

One strength of our study is that it gives a transparent and comprehensive description of the development and content of a new complex intervention for individuals with CFS. In addition, only a few qualitative studies have been published providing insight into subjective experiences with group interventions for CFS.

Conclusion and final programme

The initial programme appeared to be a feasible intervention that can be organized in a primary healthcare setting, but required modifications. The experiences of the group leaders and participants resulted in the development of the final programme (Table II). We included the eight proposed topics. Short
presentations covering these topics were made by professionals at the CFS/ME centre. In the manual for programme leaders we extended and improved the introduction and presentation of goal-setting, making action plans and problem-solving. We changed the sequences of the course elements, in line with what the participants had suggested. We also decided to add relaxation exercises because studies have shown that such exercises can have a positive effect on symptoms (39) and may improve mood, quality of life, and physical functioning (40). In addition, we included a meeting for relatives. The rest of the initial programme remained unchanged. The programme will be led by juxtaposed peer counsellors and occupational therapists who will participate in a training programme before conducting the programme. Prior to its implementation in primary healthcare settings, the effects of our final programme will be evaluated in a randomized controlled trial (20).

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References


Coping with chronic fatigue syndrome: A review and synthesis of qualitative studies

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Abstract

Background: There seems to be a need for the development of effective interventions that promote coping with chronic fatigue syndrome (CFS).

Purpose: To identify factors that promote coping with CFS and to provide recommendations for adequate interventions in this patient group.

Methods: A systematic review of qualitative studies was performed. Structured searches were conducted in major scientific databases. Two reviewers independently performed the study selection and assessment. Meta-ethnography was applied to review and synthesise the studies. The analysis provided key concepts that could be aggregated using second-order interpretations.

Results: Fifteen studies were included. The findings suggest that coping strategies, including activity management and the use of cognitive and emotional strategies, and psychological processes such as acceptance and the rebuilding of identities and lives, may promote coping with the illness. The use of adequate coping strategies appeared to be facilitated by progress in these psychological processes. Coping appeared to be facilitated mainly by self-management, occasionally complemented by treatments and social support.

Conclusions: Interventions that focus on coping with CFS may include the use of adequate coping strategies and take into account progress in psychological processes. Interventions may be complemented by self-management, other types of treatments, and social support.
Keywords

Chronic fatigue syndrome, coping, qualitative research, meta-analysis, review.

Introduction

Chronic fatigue syndrome (CFS), also known as myalgic encephalomyelitis (ME), is an illness associated with physical and cognitive disability. Living with the illness may lead to reduced quality of life, [1] identity crisis, [2] stigmatization, [3] and social isolation. [2] CFS is characterised by unexplained chronic fatigue and 4 or more of the following symptoms lasting at least six months: significant difficulties in short-term memory and concentration, sore throat, swollen lymph nodes, muscle pain, multi-joint pain, new-type headaches, sleep disturbances, and post-exertional malaise for more than 24 hours after activity. [4] The diagnosis is based on a description of symptoms, the functional level of the patient, and the exclusion of all relevant differential diagnoses. The population prevalence of CFS has been estimated to be 0.0 - 7.6 %, [5] with a higher prevalence among women. [6] To date, there is no known cure or widely accepted treatment for CFS. Some patients fully recover, whereas others become worse over time. Most patients are able to function at a stable, but reduced functional level most of the time, [7] meaning that most of the patients must learn to cope with the illness.

There exist several definitions, theories and models of coping. In the present study, coping refers to the cognitive and behavioural efforts a person makes to manage specific external and/or internal demands that are appraised as stressful. [8] According to Folkman and Lazarus, [8] these cognitive and behavioural efforts constantly change as individuals manage particular demands that are perceived as stressful. Coping processes may be directed at altering the situation that is causing distress (problem-focused coping) and/or regulating emotions as a result of a stressful condition (emotion-focused coping). [8]

To be able to cope with their illness, individuals with CFS need information, help and support from healthcare professionals who recognise their needs and provide respect and empathy. [9] Unfortunately, many patients experience trivialisation and mistrust of their illness by healthcare professionals [2,3]; consequently, they may not receive adequate help with regard to coping with CFS. Additionally, there seems to be a need for the development
of effective interventions that promote better skills for coping with the illness.

The aim of our review was to identify factors that promote coping with CFS. As we were interested in patients’ experiences with coping, this article provides a systematic review and analysis of qualitative CFS studies. Recently, two related systematic reviews concerning CFS have been conducted. Larun and Malterud [2] aimed to provide insight into patients’ and doctors’ experiences with CFS, whereas Anderson at al. [3] conducted a review of qualitative studies on CFS in general. Both studies had a broad focus, and presented some results concerning coping amongst other results. To be able to develop interventions that promote coping with the illness, we recognised a need to review the qualitative literature by focussing only on factors that promote coping with CFS. In addition, the current study differs from the two related reviews in that it includes fewer studies; however, eight of these are recent studies that were not a part of these previous reviews. By synthesising previous studies, we aimed to unite the major thematic findings of these studies and to provide interventional recommendations concerning coping with the illness.

**Methods**

Different methodologies have been presented for synthesising qualitative studies. [10] Meta-ethnography [11] is perhaps the most well developed method; therefore it was selected for this study. The meta-ethnographic synthesis of interpretive research involves the translation of studies into one another; interpretations and explanations in the original studies are translated across the studies to produce a synthesis of the studies. [11] Noblit and Hare [11] outlined a series of seven phases involved in performing a meta-ethnography (Box 1).

**Search and selection of studies**

Article selection was initiated by searching for qualitative studies on CFS published before October 2013 using the following major scientific databases: PubMed, MEDLINE, PsycINFO, AMED, EMBASE, Web of Science and CINAHL. We aimed to identify qualitative studies in English or any of the Nordic languages. The databases were searched using the following subject headings and keywords. For population, we used the following terms: fatigue syndrome, chronic; chronic/postviral fatigue disorder/syndrome; encephalomyelitis, myalgic. For coping, we used the following terms: adaptation,
psychological; coping; activities of daily living; quality of life; well-being; health knowledge/attitudes/practice; attitude to health; client attitudes; sick role; illness behaviour; coping behaviour; life experiences; emotional adjustment; self-concept; self-assessment; self-efficacy; patient satisfaction; client satisfaction; patient preferences; needs assessment; psychological needs. For methods, we used the following terms: qualitative research; interviews as topic; focus groups; narration; interview, psychological; and grounded theory. In addition three reviews associated with CFS [2,3,9] were screened to identify additional articles.

The criteria for the inclusion of studies were 1) the studies were empirical qualitative studies; 2) all patients were diagnosed with CFS by a physician/specialist or met specified criteria for CFS; 3) the majority of the participants were older than 18 years; and 4) the studies focussed on coping.

First, the studies that, broadly defined, utilised qualitative methods were identified. Second, the abstracts and titles were screened. Third, the potentially relevant articles were retrieved as full-text articles and assessed for inclusion using an include/exclude form. Finally, a quality assessment was performed according to a checklist for qualitative studies. [12] The last three steps were performed by IP and EBS independently. Differences between the reviewers’ assessments were resolved by discussion.

Analysis

The analysis began with repeated readings of the articles. In the next phase, we determined how the studies were related. We examined the articles and created a list of the reported coping concepts using the terminology from the original articles. Next, these concepts were aggregated under the emerging key concepts. By reviewing the list of concepts and key concepts, we were able to establish the relationship between the studies and aggregate them in an overarching translation (second-order interpretations).

Results

The search produced 902 articles, 121 of which utilised qualitative methods. After the titles and abstracts of these 121 studies were screened, 36 potentially relevant articles were retrieved in full text and assessed for inclusion. Eighteen articles were selected. After quality
assessment, [12] we excluded 3 studies rated as very low quality, resulting in the inclusion of 15 studies in the review. Chart 1 provides a flowchart for study screening.

The characteristics of the included studies are presented in Table 1. The review included 183 participants, the majority of whom were women. The total sample varied with respect to age and illness duration. Three of the studies included adolescents younger than 18 years. Almost all participants had been recruited from healthcare clinics, patient organisations and support groups. The majority of the studies had been conducted in the UK, whereas the other studies had been conducted in Australia, Germany, Austria, Norway, the USA, and South Africa. One study used focus group interviews to generate data, whereas the other studies used individual interviews (face-to-face or by telephone). Different analytical approaches were applied, namely, interpretative phenomenological analysis (IPA), thematic analysis, grounded theory, reconstructive interview analysis, and systematic text condensation.

Most of the studies provided information on illness severity that indicated that the review covered a range of severity. Only 4 studies reported ethnicity, with Caucasians comprising the vast majority of subjects included in these studies. [13-16] Most studies did not report socio-economic status; however, the studies that did report this information included subjects from a range of socio-economic backgrounds. [17-20]

The analysis of the included studies resulted in the identification of several concepts that could be classified into 4 emerging key concepts, namely, activity management, cognitive and emotional strategies, and psychological processes, as well as treatments and self-management, as presented in the first column in Table 2.

**Activity management**

The key concept of activity management includes pacing activities; getting sufficient rest, relaxation, and sleep; setting realistic goals for physical activity and exercise; and obtaining practical help and assistive devices.

**Pacing activities.** A number of studies indicated that pacing activities may promote better coping. Several of these studies revealed the importance of listening to body signals and monitoring physical experiences and energy levels to be able to live within limits, avoid over-exertion, and restore more rapidly [13,16-22]:

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'It’s still difficult at times, you... you want to take on more than you can deal with, but I think you learn to calm down... Yes, I do think that I’ve become more at ease, especially because I was forced to learn to become at ease. I do think that I’ve learned to... to find a balance and to listen to what my body says’. [16]

Decreasing the workload, [19,21] carefully planning activities, apportioning the available energy, [19,21] and adapting activities [15,19] were reported to be helpful coping strategies. Additionally, modifying occupations to compensate for reduced capacity, [15,21] prioritising certain tasks while eliminating others, [15,16,19,20] and avoiding specific activities, [17] as well as performing activities slowly, [19] were observed to be beneficial strategies.

**Rest, relaxation and sleep.** Balancing activities includes ensuring adequate rest. [13,15-17,19,21-23] Some studies found that the use of relaxation and meditation techniques may reduce stress levels and facilitate better rest [19,21,22]:

‘It is exceptionally boring to stay in the pool, being asked to breathe in and out. You feel as if you are 90. A Feldenkreis exercise can’t be described as any sort of action. But it works’. [19]

Moreover, resting in advance [17,19] and implementing sleep routines [14] were found to be beneficial for enhancing performance in certain occupations:

‘If I’ve got something important to do I rest and be OK for that, rather than doing something the day before’. [17]

**Realistic goals for physical activity and exercise.** Several studies revealed the perceived usefulness of setting goals for physical activity. [14,18,22-24] Some studies specified that goals might be small and realistic, [18,23] as achieving goals increased participants’ self-esteem and self-worth [18]:

‘It’s about taking baby steps and making goals em... but they have to be realistic so maybe try to cook dinner or do the food shop or something small and you feel great when you do it. You really get a boost from achieving something and you feel you have value because you cooked your wee boy’s tea or your husband comes home to a well-stocked fridge so it makes you feel better when you do something a wife should do or a Mum should do..so..yes’. [18]

Identifying barriers to reaching specific goals were also considered helpful. [24]

Larun and Malterud’s study [19] highlighted the importance of setting small and
specific goals for physical exercise, whereas Ward et al. [22] found that exercise regimes that caused participants to feel pushed to overdo it often were experienced to lead to setbacks.

**Practical help and assistive devices.** Accepting and receiving help from others with daily chores such as the housework and taking children to school was reported to be beneficial. [19,20] Additionally, having access to help and assistive devices, e.g., a wheelchair, was perceived to make it easier to pace activity level and to conserve energy for other tasks. [13]

**Cognitive and emotional strategies**

The following mental strategies were mentioned in the analysed studies: personal agency and control and dealing with emotions.

**Personal agency and control.** The importance of regaining personal agency and taking control of identifying methods of coping with the illness was revealed in most of the studies, [13-16,18-21,24] although participants expressed that gaining full control remained impossible due to problems putting advice and knowledge into practice, [14,16,24] the fluctuating nature of the illness [19,21] and on-going psychological processes with no end point [13,20]:

‘It sounds a bit daft saying that you change your life to suit CFS but if you listen to your body, monitor your condition and change your lifestyle to control this illness then you have more control’. [18]

**Dealing with emotions.** Different methods of dealing with emotions associated with the illness were described, including containing emotions and emotional threats, [20] the cultivation of an impression of health (‘passing’), [15,25] and praying/religious beliefs. [23,26] However, in most studies, the participants expressed having a positive outlook [13,20,21,23]:

‘With an illness like this... you’ve gotta try and be positive, because if you don’t [pause], there’s nothing to look forward to’. [21]

They were concentrating on ability rather than disability, [13,20] downgrading their expectations of themselves, [18-21] and comparing themselves with other people who they felt were worse off. [13,23] In addition, they expressed maintaining a hope for future
improvements, [13,20,21] discriminating between aspects of CFS that could and could not be controlled, [20] and appreciating their health, life, and relationships more than before. [23]

Psychological processes

Participants in several studies reported having experienced living through psychological processes, such as personal growth and maturity, [14] acceptance, grief, and the rebuilding of their identities and lives to be able to cope with the illness.

Acceptance. Achieving a level of acceptance appeared to be crucial to enable participants to learn to cope with their illness [13,18,20,21,27] without becoming overwhelmed and depressed by temporary setbacks [13):

‘It’s all about accepting the illness and learning to deal with it. Accepting it stops you from feeling down in the dumps and it helps you to just take each day as it comes. That helps a lot. You know that you’re going to have good days and bad days and that people don’t understand what you have but you’ve just got to get through it. There’s no point in moping around, you just have to accept it and move on. And that attitude helps you to recover. So yeah, just accept that you have it, adapt your life and move on. That’s the key because once you’ve accepted something, it’s much easier to deal with’. [18]

Reaching a stage of acceptance was described as a very difficult and painful process, [13,18,20,27] facilitated largely by time. [18,20] While Dickson et al. [18] found that personal acceptance may promote a sense of closure for participants who recognised a need to move forward with their lives, another study revealed that complete acceptance was experienced as negative and that it was important to continue to fight. [13] Acceptance was therefore considered to be an on-going and flexible process that develops and changes throughout the course of the illness. In addition to accepting the illness themselves, participants in one study mentioned the importance of accepting the actions and inactions of others associated with their condition. [27]

Rebuilding of identities and lives. The participants experienced that the illness resulted in biographical disruption, a loss of self, and a weakened identity because of the loss of future identities and identity sources, such as desired roles and activities. [18,20] Grieving was perceived to facilitate the gradual process of rebuilding identities [18,20,25,27] and involved
letting go of their old lifestyles and seeking new sources of fulfilment, e.g., engaging in new roles, activities, hobbies, or routines, to better cope with their illness [18,20,21,23]:

‘By doing a thing like this [working with a youth support group] … I’m developing another circle of friends through CFS --- it’s budding, it’s opening up again’. [20]

Some individuals reported that this process resulted in personal growth. [13,20,27]

**Treatments and self-management**

The participants in the included studies described experiences with different types of medical treatments, alternative approaches, and self-management techniques, as well as with receiving support from others to promote coping with the illness.

**Medical treatment.** Some studies reported that the participants had received rehabilitation, [19] cognitive behavioural therapy, [14,25] psycho-education [14] or counselling. [22] Dennison et al. [14] reported that participants had tried other treatments post-therapy, finding these useful in different ways and for different aspects of the illness. These treatments were usually complementary to the therapy they had received.

Positive reactions were expressed toward healthcare professionals who were interested in CFS and who recognised and validated the condition [14,25]:

‘Relief in the facts that they believed me, that they accepted that there is something there. The worse thing about it is the not being believed, like you’re being ignored, like you’re being treated like an idiot’. [25]

Additionally, participants expressed that it was important to encounter healthcare professionals who listen, understand, and offer appropriate challenges [22] and emotional support [14]:

‘… when the counsellor is truly empathic with you, it’s at that moment when there’s a release’. [22]

Negative reactions that were observed were feeling pigeonholed and being subject to generalisations, [14] being wrongly categorised as someone with predominantly mental or psychological problems rather than with physical health problems, [14,22] and undergoing counselling that was perceived as controlling or patronising or as a form of brainwashing. [22]
Brooks et al. [25] found that a lack of confidence in healthcare professionals may have clear implications for patients’ management of their condition.

**Alternative approaches.** Participants in several studies reported that they had tried alternative techniques such as distant healing, [26] the Lightning Process, [24] acupuncture, dietary supplements, [13,23] homeopathy, kinesiology, osteopathy, Reiki and spiritual healing, [13] and described both positive and negative experiences with such approaches. Reme et al. [24] found that it was perceived to be important for staff to be positive and encouraging. Moreover, value was assigned to receiving support from other sufferers who were participating in the programme, leading to engagement and treatment commitment [24]:

‘It was useful being in a group as well, getting to see other people walking through it and other people sort of answering their questions was useful’. [24]

**Self-management.** Although some studies reported experiences with medical or alternative treatments, many studies [15,16,18,21,27] did not report that subjects had received any type of help from healthcare professionals at all. Therefore, many sufferers reported that they had to learn how to cope with the illness by themselves:

‘I think I had to find a lot of it out for myself, how to deal with it, how to handle it’. [13]

Self-management techniques that were described were gaining knowledge [13,20,21] from other sufferers, [13,21] and the media, [13] as well as experiential learning [13,18-21] and self-reflection. [13,18,20,21,27] The use of self-management techniques was also mentioned by participants who had received medical or alternative treatments both before and after receiving treatment.

Receiving concrete and individually adapted advice from others was considered to be helpful, especially when the advice supported the need to establish a balance between rest and exercise. [19,23] Obtaining advice from other sufferers was also found to be useful, as it may lead to better understanding of their illness. [13,21,24]

**Social support.** The establishment of safe relationships was one of the strategies participants used to protect themselves against threats associated with CFS. [20,25] One study revealed that patients felt it was important to receive understanding and support from other individuals who were able to continue to acknowledge and validate the ‘real’ person (i.e. the healthy and active individual before the illness). [25] Young people expressed that they needed their parents for emotional support and that it was important for them that parents were involved in
the implementation of advice and strategies and the enforcement of rules. [14] Although it was important to receive support, participants expressed a need for ‘space’ to be alone as well. [23]

Second-order interpretations

After reviewing the list of concepts and key concepts, it became clear that the aggregated studies represented a ‘line of argument’ and could be assembled into an overarching translation (second-order interpretations), [11] as presented in the second column of Table 2.

The use of adequate coping strategies appears to be facilitated by progress in psychological processes. Several studies described psychological processes [13,18,20,21,25,27] that appeared to influence the use of adequate coping strategies. Non-acceptance, biographical disruption, a loss of self and a weakened identity may promote the use of unhelpful coping strategies, such as ignoring their limits, resulting in setbacks [13] and social withdrawal, [18,20,27] as well as compromised perceptions of self-agency and control. [18,20] On the other hand, acceptance and the rebuilding of identities and lives may promote the use of more adequate coping strategies. [13,18,20,21,27] The described psychological processes do not appear to be unidirectional; rather, acceptance and the rebuilding of identities and lives were found to fluctuate along a continuum over time. [13,20,25,27] Backward movements were perceived to occur because of the chronic and fluctuating nature of the illness [13,20,27] and exposure to negative social constructions of CFS. [20]

Coping with CFS appears to be facilitated mainly by self-management, occasionally complemented by treatments and social support. The included studies described how the use of self-management techniques may facilitate both the use of adequate coping techniques [13,15,16,18-21] and progress in the described psychological processes. [13,18,20,21,27] Self-management was occasionally complemented by medical treatment, [14,19,22,25] alternative approaches, [13,23,24,26] and social support, [13,14,19,23,24] mainly facilitating the use of adequate coping strategies without referring to psychological processes.

Discussion and conclusion
In this study, meta-ethnography, as described by Noblit and Hare, [11] was used to synthesise the results of qualitative research. Based on the findings in the included studies, we developed second-order interpretations, which were at the same time consistent with the original results and also extended beyond them.

Our synthesis indicated that the use of adequate coping strategies, such as activity management (problem-focused coping) and mental strategies (emotion-focused coping), may be facilitated by progress in psychological processes. Our findings were consistent with findings from other studies presenting theories on coping with different chronic illnesses. [28-30] However, listening to body signals and monitoring physical experiences and energy levels to enable individuals to live within their limits, preventing over-exertion and enabling faster restoration, appear to be more typical coping strategies for individuals with CFS. This finding is not surprising, as even minor over-exertions may lead to extensive setbacks. [13,16,31,32] The usefulness of pacing, however, is disputed among scientists, as quantitative studies have shown conflicting results regarding interventions including pacing. [33,34] Slightly divergent interpretations of pacing in these studies may explain the differences in outcomes, [35] as practising pacing by under-expending available energy may not be beneficial. [36].

Our findings were consistent with the results of other reviews that indicated that many individuals with CFS must learn how to cope with the illness by themselves without perceiving adequate information, help and support from healthcare professionals. [2,3,9] This may explain why participants in several studies had tried several alternative approaches, as found by Featherstone. [37] However, research has provided limited evidence for the effectiveness of alternative approaches in relieving symptoms of CFS. [38]

Interestingly, the included studies in our review that focused on the perceived benefits of different types of treatments did not refer to psychological processes, while our review revealed that the use of adequate coping strategies appeared to be facilitated by progress in psychological processes. Therefore, interventions for individuals with CFS may not only focus on the use of adequate coping strategies but also on psychological processes to promote coping. Because acceptance and the rebuilding of identities and lives were found to fluctuate along a continuum over time, [13,20,25,27] interventions may be flexible and take into account individual movements along these continuums that can hinder or facilitate the use of adequate coping strategies at certain times. Our findings may indicate that a good relationship with healthcare professionals can have implications for the management of the illness. [25] Thus, interventions may be provided by professionals who understand, recognise and validate the CFS condition as well as offer emotional support and appropriate challenges.
Chew-Graham et al. [39] found that acceptance of the diagnosis can influence whether or not a patient engages in a treatment, while our review found that the use of adequate coping strategies may be influenced by acceptance of the illness which means not only accepting the diagnosis but also accepting being ill.

In addition, Chew-Graham et al. [39] found that engagement in an intervention can be influenced by the extent to which the content of the intervention matches the patient’s illness beliefs. Our recommendations are based on patient’s experiences with coping. Therefore, we expect that interventions based on our recommendations will be acceptable for CFS patients. Our review has some limitations. Meta-ethnography has been demonstrated to be a useful approach [40] and has been used in several studies. [2,3,40,41]. However, many aspects of the phases in the synthesing process remain ill-defined, [42] resulting in practical challenges [42] and different interpretations of the approach. [2,3,40]

One of our challenges was locating relevant studies. Our research strategy was not able to detect all potentially eligible studies, as two of the included studies [16,23] were identified through other sources. We may also have been unable to identify potentially eligible qualitative studies that were published after May 2010. [3]

Another challenge was inclusion decisions. Although we attempted to include studies that focussed on similar populations, different CFS case definitions were utilised in each study, which may have contributed to differences in findings in the included studies and may have influenced the overall synthesis. Additionally, many of the studies may be biased, as the vast majority of participants were recruited from healthcare clinics, patient organisations and support groups, and almost all studies were conducted in Western countries. The population in this review may therefore not be representative of all individuals with CFS.

The assessment of quality in qualitative research has been subject to debate. [43,44] We decided to perform a quality assessment to enable the exclusion of studies with very low methodological quality to avoid distortion of the overall synthesis. [44]

Both qualitative and quantitative studies are needed to evaluate the experiences with and the effects of interventions that are based on our recommendations. Self-management, other types of treatments, and social support may complement interventions that focus on coping with CFS.
Acknowledgements

We would like to thank Marie Isachsen, librarian at Oslo University Hospital, for providing valuable support during literature search. We also would like to thank Lillebeth Larun, Ph.D., at the Norwegian Knowledge Centre for the Health Services, for useful advice regarding the initial preparations of this study.

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Declaration of Conflicting Interests

The authors declare that there is no conflict of interest.
References


1. Getting started
2. Deciding what is relevant to the initial interest
3. Reading the studies
4. Determining how the studies are related
5. Translating the studies into one another
6. Synthesising translations
7. Expressing the synthesis

**Box 1.** The seven phases of Noblit and Hare’s meta-ethnography (11)
Articles identified through database searching (n = 894)

Additional articles identified through other sources (n = 8)

Articles screened for use of qualitative methods (n = 902)

Articles excluded (n = 781)

Screened articles that utilized qualitative methods (n = 121)

Articles excluded (n = 85)

Full-text articles assessed for eligibility (n = 36)

Articles excluded (n = 18)

Articles assessed for quality (n = 18)

Articles excluded (n = 3)

Articles included in qualitative synthesis (n = 15)

**Chart 1.** Identification and selection of qualitative studies
<table>
<thead>
<tr>
<th>Study</th>
<th>Population (females)</th>
<th>Age in years (mean)</th>
<th>Illness duration in years (mean)</th>
<th>Diagnosis Case definition</th>
<th>Recruitment (country)</th>
<th>Methods/Analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Arroll and Howard (28)</td>
<td>N = 10 (7)</td>
<td>(39.5)</td>
<td>(7.4)</td>
<td>Physician/specialist Fukuda</td>
<td>ME/CFS support groups and personal contacts (UK)</td>
<td>Individual semi-structured interviews/ interpretative phenomenological analysis (IPA)</td>
</tr>
<tr>
<td>Brooks et al. (26)</td>
<td>N = 2 (0)</td>
<td>Midt-fifties</td>
<td>13-25 (19)</td>
<td>Physician/specialist Fukuda</td>
<td>Hospital immunology clinic (UK)</td>
<td>Individual semi-structured interviews/IPA</td>
</tr>
<tr>
<td>Clements et al. (18)</td>
<td>N = 66 (46)</td>
<td>18-56 (36)</td>
<td>0.5-7 (2.25)</td>
<td>Physician/specialist Fukuda and Oxford</td>
<td>Medical infectious disease clinic (UK)</td>
<td>Individual semi-structured interviews/thematic</td>
</tr>
<tr>
<td>Dennison et al. (15)</td>
<td>N = 16 (10)</td>
<td>16-24 (19.9)</td>
<td>Not stated</td>
<td>Physician/specialist Fukuda or Oxford</td>
<td>Hospital outpatient clinic (among participants in a treatment trial) (UK)</td>
<td>Individual semi-structured telephone interviews/thematic</td>
</tr>
<tr>
<td>Dickson et al. (19)</td>
<td>N = 14 (8)</td>
<td>21-68 (not stated)</td>
<td>Not stated</td>
<td>Physician/specialist Fukuda</td>
<td>Alternative therapy clinic and personal contacts (UK)</td>
<td>Individual semi-structured interviews/IPA</td>
</tr>
<tr>
<td>Edwards et al. (14)</td>
<td>N = 8 (8)</td>
<td>37-55 (42.5)</td>
<td>1.5-12 (5.9)</td>
<td>Physician/specialist Not specified</td>
<td>ME self-help network (UK)</td>
<td>Individual semi-structured interviews/IPA</td>
</tr>
<tr>
<td>Gray and Fossey (22)</td>
<td>N = 5 (not stated)</td>
<td>16-44 (not stated)</td>
<td>2-10 (not stated)</td>
<td>Physician/specialist Not specified</td>
<td>Patient organisation newsletter (Australia)</td>
<td>Individual semi-structured interviews/thematic</td>
</tr>
<tr>
<td>Güthlin et al.</td>
<td>N = 17 (13)</td>
<td>31-66 (not stated)</td>
<td>Not stated</td>
<td>Physician/specialist</td>
<td>Private practices for</td>
<td>Individual semi-structured interviews/thematic</td>
</tr>
<tr>
<td>Study</td>
<td>Sampling Method</td>
<td>Sample Size</td>
<td>Age Range (Mean)</td>
<td>Referrals</td>
<td>Setting</td>
<td>Data Collection Method</td>
</tr>
<tr>
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</tr>
<tr>
<td>Fukuda and Oxford (20)</td>
<td>Environmental medicine (among participants in a healing trial) (Germany and Austria)</td>
<td>N = 10 (8)</td>
<td>40-64 (50)</td>
<td>Not specified</td>
<td>Rehabilitation clinic (Norway)</td>
<td>Telephone interviews/reconstructive interview analysis</td>
</tr>
<tr>
<td>Makarud (20)</td>
<td>Not stated</td>
<td>N = 4 (4)</td>
<td>20-50</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Semi-structured focus group interviews/systematic text condensation</td>
</tr>
<tr>
<td>Lombard and Mouton (17)</td>
<td>Not stated</td>
<td>N = 12 (5)</td>
<td>27-61 (40.33)</td>
<td>Not stated</td>
<td>Physician and personal referrals, existing lists of CFS sufferers and media advertisements (South Africa)</td>
<td>Individual semi-structured interviews and autobiographical sketches/ thematic analysis</td>
</tr>
<tr>
<td>Lovell (24)</td>
<td>Not stated</td>
<td>N = 9 (8)</td>
<td>14-26 (not stated)</td>
<td>Not specified</td>
<td>Travel health clinic for overseas aid workers (UK)</td>
<td>Individual semi-structured interviews/grounded theory</td>
</tr>
<tr>
<td>Reme et al. (25)</td>
<td>Not stated</td>
<td>N = 19 (14)</td>
<td>20-75 (49)</td>
<td>Not specified</td>
<td>Website patient organisation (UK)</td>
<td>Individual semi-structured telephone interviews/grounded theory</td>
</tr>
<tr>
<td>Travers and Lawler (21)</td>
<td>Not stated</td>
<td>N = 10 (8)</td>
<td>20-75 (49)</td>
<td>Not specified</td>
<td>CFS newsletter, brochures and referrals (Australia)</td>
<td>Individual semi-structured interviews/grounded theory</td>
</tr>
<tr>
<td>Ward et al. (23)</td>
<td>N = 25 (21)</td>
<td>23-65 (44)</td>
<td>2-19 (not stated)</td>
<td>Physician/specialist Not specified</td>
<td>Patient organisation newsletter (UK)</td>
<td>Individual unstructured telephone interviews/thematic</td>
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</tr>
<tr>
<td>Ware (16)</td>
<td>N = 66 (53)</td>
<td>27-72 (43)</td>
<td>2.5-36 (not stated)</td>
<td>Not specified Holmes</td>
<td>Research centre and women’s hospital clinic (USA)</td>
<td>Individual semi-structured interviews/thematic</td>
</tr>
</tbody>
</table>

**Table 1.** Features of the qualitative studies included in the analysis
<table>
<thead>
<tr>
<th>Concepts</th>
<th>Second-order interpretations</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Activity management:</strong> pacing activities,</td>
<td>The use of adequate coping strategies appears to</td>
</tr>
<tr>
<td>rest, relaxation and sleep, realistic goals,</td>
<td>be facilitated by progress in psychological processes</td>
</tr>
<tr>
<td>practical help and assistive devices</td>
<td></td>
</tr>
<tr>
<td><strong>Cognitive and emotional strategies:</strong></td>
<td>Coping with CFS appears to be facilitated mainly by self-management, occasionally</td>
</tr>
<tr>
<td>personal agency and control, dealing with</td>
<td>complemented by treatments and social support</td>
</tr>
<tr>
<td>emotions</td>
<td></td>
</tr>
<tr>
<td><strong>Psychological processes:</strong> acceptance,</td>
<td></td>
</tr>
<tr>
<td>rebuilding of identities and lives</td>
<td></td>
</tr>
<tr>
<td><strong>Treatments and self-management:</strong></td>
<td></td>
</tr>
<tr>
<td>medical treatment, alternative approaches,</td>
<td></td>
</tr>
<tr>
<td>self-management and social support</td>
<td></td>
</tr>
</tbody>
</table>

Table 2. Synthesis, including concepts and second-order interpretations
Effectiveness of a group-based self-management program for people with chronic fatigue syndrome: A randomized controlled trial

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Abstract

Objective: To evaluate the effectiveness of a group-based self-management program for people with chronic fatigue syndrome (CFS).

Design: A randomized controlled trial.

Setting: Four mid-sized towns in southern Norway and two suburbs of Oslo.

Subjects: A total of 137 adults with CFS.

Intervention: A self-management program including eight biweekly meetings of 2.5 hours duration. The control group received usual care.

Main measures: Primary outcome measure: MOS-SF-36 physical functioning subscale. Secondary outcome measures: fatigue severity scale, self-efficacy scale, physical and mental component summary of the MOS-SF-36, and the illness cognition questionnaire (acceptance subscale). Assessments were performed at baseline, and at six-month and one-year post-intervention.

Results: No significant differences were found within or between the groups in physical functioning. At the six-month follow-up, a significant difference between the two groups was found concerning fatigue severity (p = 0.039) in favor of the control group, and concerning self-efficacy in favor of the intervention group (p = 0.039). These significant differences were
not sustained at the one-year follow-up. Acceptance improved significantly in the intervention
group at the six-month (mean change = 0.9, p = 0.006) and one-year (mean change = 0.7, p =
0.030) follow-ups and in the control group at the six-month follow-up (mean change = 1.1, p
= 0.001). The dropout rate was low (13.9 %) and the median number of sessions attended was
high (seven of maximum eight).

**Conclusions:** Long-term follow-up studies are needed before firm conclusions can be drawn
on the effectiveness of the intervention.

**Keywords**
Chronic fatigue syndrome, randomized controlled trial, patient education, coping, self-
efficacy

**Introduction**

Chronic fatigue syndrome (CFS), also known as myalgic encephalopathy (ME), is a complex
condition defined by six months or more of unexplained fatigue and a range of additional
cognitive and physical symptoms (1). The illness may lead to substantial reductions in
occupational performance (2) and health status (3).

CFS has no known cause. Because there are no specific diagnostic tests, the diagnosis
is based on the description of symptoms, the functional level of the patient and the exclusion
of alternative diagnoses. The prevalence of CFS is estimated to be 0.2 – 0.4 % of the
population (4), with a higher prevalence among women (5). Currently, there is no cure or
widely accepted treatment for CFS. Although some patients recover, CFS remains a chronic,
debilitating condition for the majority of the patients (6, 7), meaning that these patients must
learn how to cope with the illness.

Studies have shown that coping with CFS can be improved by activity management,
such as pacing activities (8-11), ensuring adequate rest (9-11), and access to practical help and
assistive devices (10, 12), as well as personal agency (8-10, 12) and having a positive outlook
(9, 11, 12). In addition, improved coping appears to depend on the progress in psychological
processes, such as grief, acceptance, and the rebuilding of identities and lives (8, 12).

In spite of the negative effects of the illness, several studies have shown that patients
receive insufficient information, support and help from their general practitioners (GPs) and
other healthcare professionals after diagnosis (9, 13), although recent research has revealed
that receiving help at an early phase of the illness to cope with CFS, prevents relapses and
results in a better prognosis (14).

In Norway, the structure of the healthcare system is guided by the Coordination Reform, which promotes access to good and equal healthcare services, regardless of one’s place of residence. This reform emphasizes ‘coping’ and the need for establishing patient education programs in primary healthcare as one of several potential actions (15). Some studies have shown that patient education programs appear to be beneficial for people with CFS (16, 17). However, several studies have indicated that patient education alone is not sufficient to improve coping (18, 19) and should be complemented by self-management education (20). A self-management program allows patients to identify their problems and provides techniques for helping patients make decisions, take appropriate actions, and alter these actions as they encounter changes in their circumstances or during the course of their illness (20). The CFS Empowerment Project, which was conducted in the United States of America, included a self-management program for CFS patients that showed positive effects regarding symptom severity (21), quality of life (21, 22), and resource acquisition (23).

None of these programs for CFS patients could be implemented in the Norwegian healthcare system without modifications, because of the specific organization of healthcare, and limited access to healthcare professionals with competence in CFS in primary healthcare. Therefore, a pilot study was conducted to develop a self-management program that could be organized in a primary healthcare setting in Norway (unpublished). The results of this pilot study were used in the development of the final program that was evaluated in the current study. The pilot study indicated that participation in this program might lead to increased acceptance of the illness. Several other studies concerning CFS have found an association between the acquisition of knowledge and improved understanding and acceptance (24). Studies concerning CFS have revealed that understanding and accepting the illness seem to facilitate coping (9, 11), as acceptance may promote the downgrading of expectations and the search for new sources of fulfillment (9, 11, 13). In addition, Van Damme and colleagues determined that acceptance had a positive effect upon fatigue and psychological aspects of well-being (25).

Our self-management program was based on self-efficacy theory as self-efficacy perceptions play an important mediating role in self-management activities (26). In the context of social cognitive theory, where personal attributions are mediators of behavior (27), Bandura states that ‘perceived self-efficacy refers to beliefs in one’s capabilities to organize and execute the courses of actions required to produce given attainments’ (27, page 3). Self-efficacy specifically implicates personal judgments of performance capabilities in a given
domain of activity (27). Consequently, our program focused on regaining control in relation to CFS complains. It has been shown that self-management programs for people with chronic conditions based on self-efficacy theory may lead to improvements in health status and self-efficacy in people with chronic illnesses (28-30). To our knowledge there are no publications addressing interventions based on self-efficacy theory for individuals with CFS.

Pacing was a central topic in our program. Several qualitative studies have revealed that people with CFS perceived pacing as a useful coping strategy (8-11). These findings are consistent with the results of a survey sponsored by the ME Association in the UK (31). The usefulness of pacing, however, is disputed among scientist, as quantitative studies have shown conflicting results regarding interventions including pacing (32-34). Slightly divergent interpretations of pacing in these studies might explain the differences in outcomes (35). Therefore, pacing in our program was based on the Energy Envelope Theory. This theory is based on the hypothesis that avoiding over-exertion by maintaining expended energy levels within the envelope of perceived energy levels, might prevent relapses and increase energy levels and the tolerance of activity (36, 37). Several studies concerning CFS patients have shown results supporting the Energy Envelope Theory (34, 38).

The aim of this study was to evaluate the effectiveness of our group-based self-management program in a randomized controlled trial. Based on prior research, we formulated two hypotheses. Our primary hypothesis was that the intervention would lead to improvements in physical functioning for people with CFS, as compared to people with CFS receiving usual care. The secondary hypothesis was that the intervention would lead to improvements in fatigue severity, self-efficacy, acceptance and health status.

Methods

Study design
This randomized controlled trial had two arms: an intervention group that was participating in the self-management program and a control group that was offered participation in the self-management program after completing the final outcome measures. Participants in the control group were allowed to receive treatment as usual, but they were excluded from participation in the patient education program at our hospital. Evaluations were carried out at baseline, and at six-month and one-year post-intervention.

The study was approved by the Privacy Ombudsman for Research at our hospital and
the Regional Committee for Medical Research Ethics (Health Region North, approval number 2011/894/REK nord).

**Participant recruitment and initial screening**

Occupational therapists and peer counselors in four mid-sized towns in southern Norway and in two suburbs of Oslo were willing to conduct the self-management program. Participants were recruited from these towns and suburbs and from the surrounding municipalities from November 2011 until January 2012. The participants were recruited from a variety of sources, including healthcare professionals, waiting lists for the patient education program at our hospital and CFS patient organizations. Information regarding the study was disseminated through brochures and personal communication. In addition, study announcements for participants were placed on the website of our hospital.

Participants were required to be older than 18 years, to be diagnosed with CFS by a physician or medical specialist, to meet the Centers for Disease Control and Prevention (CDC) research diagnostic criteria (1) and the research or clinical Canadian diagnostic criteria (39), to not be pregnant, to be able to read and speak Norwegian, and to be considered to be physically able to attend the program. The patients who were interested in participating in the study were given additional information by telephone. They were asked to complete and return a consent form and the DePaul symptom questionnaire (DSQ) (40).

The (DSQ) (40) is a self-report measure that has items that measure the dimensions of the CDC CFS (1) and the Canadian ME/CFS (39) case definitions by rating the frequency and severity of 54 symptoms over the past six months on a five-point scale. The DSQ has been shown to have good test-retest reliability among patient and control groups (41). The initial screening was supplemented by a baseline registration of over-exertion. The participants were asked to rate perceived energy and expended energy over the past week on a 100-point scale, with 0 = no energy and 100 = abundant energy similar to when the person was completely well. Perceived energy referred to the participants’ estimation of their available energy resources, whereas expended energy was defined as the participants’ estimation of the total amount of energy exerted. Expended energy could be greater than perceived energy when participants were pushing themselves over their energy limits. The participants’ expended energy was divided by their perceived energy, and this number was then multiplied by 100. A score of equal to or below 100 is considered to be remaining within one’s energy envelope (42); however, a cut off score of 150 has also been used (34). Both of these cut off scores were used in our study.
The written informed consent we obtained from the participants gave us permission to request confirmation regarding their CFS-diagnoses from their physicians or medical specialists.

**Intervention**

The self-management program was conducted by one juxtaposed peer and occupational therapist after participation in a three-day training program. They followed a detailed manual to teach the self-management program. The program was conducted in the spring of 2012 with 6 – 14 participants in each group and included eight biweekly meetings of 2.5 hours duration. The participants were taught how to take greater initiative for coping with their illness and for dealings with healthcare professionals and significant others. These capabilities were developed through educational presentations, the exchange of experiences among participants, modeling of self-management skills, guided mastery practice, and informative feedback (27). The content of the program is presented in Table 1. The program covered a number of topics relevant to coping with CFS. At the beginning of the program, the participants set personal goals for coping and constructed action plans to achieve these goals. During each meeting, action plans were evaluated, and the participants were offered feedback from the other participants in overcoming obstacles they faced. At the end of each meeting, the current action plans were adjusted if necessary or were replaced with new action plans. In addition, one meeting was organized for relatives of the participants with an educational presentation about CFS; this meeting covered the content of the self-management program and allowed for the exchange of experiences among relatives.

**Outcome measures**

We performed assessments at baseline, and at six-month and one-year follow-ups. One-year follow-up measurements were performed to evaluate whether the changes found at the six-month follow-up were sustained at the one-year follow-up. The outcome measures were collected using questionnaires that were completed at home and returned by mail.

*Physical functioning:* The Medical Outcomes Study 36-item Short Form Questionnaire (SF-36) is a health survey with scales for physical functioning, role-physical functioning, bodily pain, general health, vitality, social functioning, role-emotional functioning, and mental health (43). The physical functioning subscale, served as the primary outcome measure (range 0 - 100, with higher scores denoting better levels of functioning). This subscale has been used as a primary outcome measure in several other CFS studies (32, 33). The SF-36 has been validated in several international and national studies and has
demonstrated adequate psychometric properties as a measure of functional status in a CFS population (44).

We also utilized some secondary outcome measures, as follows.

**Fatigue:** The fatigue severity scale (FSS), developed by Krupp and colleagues (45), was used for measuring fatigue severity, which is the main symptom in CFS (range 9 – 63, with higher scores indicating higher levels of fatigue severity). The FSS has demonstrated adequate psychometric properties (46) and was found to be appropriate for measuring fatigue in people with CFS (47). This measure has been used in several CFS studies (32, 48).

**Self-efficacy:** The self-efficacy scale (49) measured the sense of control in relation to CFS complaints. The ratings from each of the five questions were added (range 5 – 24), and higher scores reflect higher levels of self-efficacy. Cronbach’s alpha reliability has ranged from 0.70 to 0.77 (49). The scale has been used in several CFS studies (32, 49).

**Acceptance:** The illness cognition questionnaire (ICQ) measures illness cognitions in patients with chronic diseases (50). The ICQ consists of 18 items. For the current study, the acceptance subscale (six items) was applied (range 6 – 24, with higher scores indicating higher levels of acceptance). Research has indicated that the ICQ is a reliable and valid instrument (50). The ICQ has been used in CFS studies (25, 51).

**Health status:** The eight scales of the SF-36 (43) can be aggregated into two summary measures based on weighted sums of the scale scores. The two summary measures are the physical component summary (PCS) for physical health and the mental component summary (MCS) for mental health (52). Because some discrepancies between the summary scores and the scores on the individual scales have been observed (53), these problems were reduced by using oblique scores. The PCS and MCS oblique scores were computed using an online calculator provided by the SF-36 Community (http://www.sf-36.org) and are based on Norwegian reference data. PCS and MCS scores below 40 points indicate poor health (54). This measure has been used in CFS studies (3).

**Sample size calculation**

In a previous publication (55), we found that to obtain 80 % power with a 5 % significance level, a minimum of 56 participants must be included in each group to detect a difference of 15 points (SD = 28) on the SF-36 of subscale physical functioning between the groups. We expected a dropout rate of approximately 25 % (32); therefore, we planned to include 75 participants in each group.
Randomization and blinding

After enrollment into the study, the participants were randomly assigned to the intervention or control group. Random assignment was done using a computerized randomization list that was created in blocks of six with the municipality as the stratifying variable (56). The assessor was blinded to the group allocation of the participants.

Statistical analysis

SPSS, version 15 (SPSS Inc., Chicago, IL, USA) was used for the statistical analyses. At baseline, the independent-samples t-test was used when comparing the primary and secondary outcomes between the groups. In addition, the Mann-Whitney U-test was used for comparing continuous variables with a skewed distribution, whereas the chi-squared test was used for comparing binary variables. The paired-samples t-test was used for within-group comparisons between baseline and the six-month and one-year follow-ups. Analyses were carried out as “per-protocol” analyses, where only participants with complete registrations on an outcome measure were included in the statistical analyses of this measure. A significance level of five percent was used.

To investigate the effect of the intervention on physical functioning, fatigue severity, self-efficacy, acceptance and health status, as compared to controls, a mixed model analysis for repeated measures was performed. Time of measurement (baseline, and six-month and one-year follow-ups) was used as a within-subjects factor and group (intervention or control) as a between-subjects factor.

When less than 50 % of the values on a subscale were missing, the values were replaced by using an imputation procedure in SPSS. A total of 134 values (0.003 %) of 47,012 possible values were imputed.

Results

Participant flow

The participant flow is presented in Figure 1. A total of 146 included participants were randomly assigned to the intervention or control group with 73 participants in each group. After randomization, nine participants withdrew from the study; two withdrew from the intervention group because they were too ill to participate, and seven withdrew from the control group because they were too ill (1), did not accept randomization (1) or for an unknown reason (5). A total of 137 participants remained in the study at baseline, and 118
completed the assessments after one year. Therefore, the total dropout rate was 13.9 %. The median number of sessions attended was seven. Five participants attended 0 - 2 sessions because they were too ill to participate (4) or because they moved to another town (1). The remaining 66 participants attended 4 - 8 sessions each.

**Baseline characteristics**

The baseline characteristics are presented in Table 2. A significant difference between the intervention and control group was found only for gender (p = 0.022).

**Effect on outcome measures**

The outcomes at the six-month follow-up and at the one-year follow-up were compared with the baseline values. The scores on the outcome measures are presented in Tables 3a and 3b. Analyses of the entire sample showed no significant differences between men and women on any of the outcome measures.

**Primary outcome.** No significant differences were found within or between the groups on the SF-36 physical functioning subscale at any of the measure points.

**Secondary outcomes.** At the six-month follow-up, a significant difference between the two groups was found concerning fatigue severity (p = 0.039) in favor of the control group. This difference between the groups was not sustained at the one-year follow-up.

The control group showed significant decreases in self-efficacy at the six-month follow-up (mean change = -0.8, p = 0.044), and a significant difference between the groups was found at the six-month follow-up in favor of the intervention group (p = 0.039). This difference between the groups was not sustained at the one-year follow-up.

Acceptance improved significantly in the intervention group at the six-month (mean change = 0.9, p = 0.006) and one-year (mean change = 0.7, p = 0.030) follow-ups and in the control group at the six-month follow-up (mean change = 1.1, p = 0.001). There were no significant differences between the two groups at any of these measure points.

No significant differences were found within or between the groups concerning health status at any of the measure points.

Because five participants in the intervention group attended two or fewer sessions, an additional statistical analysis was conducted without these five participants. This analysis revealed similar results (data not shown).
Discussion

The present trial showed significant differences between the two groups concerning fatigue severity in favor of the control group and concerning self-efficacy in favor of the intervention group at the six-month follow-up. These significant differences were not sustained at the one-year follow-up. The intervention had no effect on physical functioning, health status or acceptance for people with CFS, as compared to people with CFS receiving usual care.

Because pacing in our program was based on the Energy Envelope Theory, it was expected that participation in the program would lead to improvements in physical functioning (34, 38); however, the intervention group reported unchanged physical functioning. The lack of improvement in physical functioning may be explained by the fact that the majority of the participants in our study already was able to stay within their ‘energy envelope’ at baseline (Table 1) and, consequently, might have experienced improvements in physical functioning prior to participation in the program. On the other hand, Brown and colleagues (42) determined that those patients who were closer to remaining within their ‘energy envelope’ were more impaired in physical functioning, than those who were moderately overextending. This may indicate that the use of more adaptive strategies is not associated with improvements in physical functioning (42). Our program encouraged increasing activity levels when the participants were able to manage their daily activities, and symptom fluctuations were reduced to a manageable level. The lack of improvement in physical functioning in the intervention group may indicate that the participants continued to operate at lower levels than advocated. The use of an objective measure of activity might have confirmed compliance.

Based on the results of prior studies using pacing according to the Energy Envelope Theory, it was also expected that the program would lead to improvements in fatigue (34, 38). Throughout the whole study, the intervention group reported unchanged fatigue severity, whereas the control group reported improved fatigue severity. A significant difference between the two groups was found at six-month follow-up in favor of the control group. This is a surprising result and may indicate that remaining within the ‘energy envelope’ is not associated with improvements in fatigue either, as found for physical functioning (42).

Similar studies have found improvements in self-efficacy (28, 29, 32, 57); however, we did not find improvements in self-efficacy in the intervention group. Moreover, it was surprising that the control group showed a significant decline in self-efficacy at the six-month follow-up. In addition, we found a significant difference between the groups at six-month
follow-up in favor of the intervention group, indicating that participation in the self-management program may have prevented a decline in self-efficacy. This result may indicate that the individuals in the control group did not receive adequate help and support from health professionals to be able to cope with their illness (13). That the difference between the groups was not sustained at one-year follow-up may indicate a need for follow-up over a longer time period, as offered in the CFS Empowerment Project (21).

Several studies have found an association between the acquisition of knowledge and improved understanding and acceptance (24). Because exchange of information and experiences was part of the intervention program, the intervention group showed significant improvements in acceptance at six-month and one-year follow-ups, as expected. This is a positive result as studies concerning CFS have revealed that accepting the illness seems to facilitate coping (9, 11). However, it was surprising that we did not find differences in acceptance between the groups.

Van Damme and colleagues (25) found a positive relationship between acceptance and psychological aspects of well-being. In our study, however, both groups reported unchanged health status, indicating that changes in health status do not necessarily occur immediately after improvements in acceptance.

Our study had several limitations. It should be noted that our program lasted for only 15 weeks, partly based on experiences with self-management programs for people with chronic illnesses (28). Participants in other trials concerning CFS were seen more often and were followed for a longer period of time (21, 32, 33). Whiting and colleagues (58) determined that CFS studies including a longer treatment duration were more likely to report positive effects from the intervention, which may explain the lack of improvements in our study in terms of physical functioning and health status.

Our program included group sessions alone, whereas other interventions for CFS typically include only individual consultations (32, 33) or a combination of group interventions and individual consultations (21). Our group intervention might have achieved better outcomes if we had also included individual consultations (27).

Individuals with CFS comprise a heterogeneous group; therefore, subgrouping of individuals has been recommended. In our study, it would have been interesting to evaluate whether the self-management program was more effective for those who were overextending. However, due to the relatively small sample size, subgrouping of individuals based on their particular cluster of symptoms was not appropriate for our study.

The low dropout rate and the high number of sessions attended may indicate that this
program is an acceptable and helpful intervention. This may raise the question to what extend the outcome measures that were applied in this study could detect the effects of the intervention.

Our results were obtained from a large randomized controlled trial with a low dropout rate (32) and a final measure point at the one-year follow-up. This study design is supposed to strengthen the reliability of our results.

In conclusion, this study suggests that the evaluated self-management program for people with CFS had a stabilizing effect on fatigue severity and self-efficacy for people with CFS at the six-month follow-up, as compared to people with CFS receiving usual care. All participants showed improvements in acceptance. Longer-term quantitative and qualitative follow-up studies are needed before we can draw firm conclusions on the effectiveness of our self-management program.

Acknowledgements

We would like to thank the study participants and the group leaders for their participation.

Conflicts of Interest Statement

We are not aware of any conflicts of interest.

Funding

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53. Farivar SS, Cunningham WE and Hays RD. Correlated physical and mental health summary scores for the SF-36 and SF-12 Health Survey, V.I. *Health Qual Life Outcomes* 2007; 5: 54.


**Clinical messages**

- The self-management program had a stabilizing effect on fatigue severity and self-efficacy at the six-month follow-up.
- All participants showed improvements in acceptance.
- Longer-term follow-up studies are needed before we can draw firm conclusions on the effectiveness of the self-management program.
Table 1: Content of the program

<table>
<thead>
<tr>
<th>Day</th>
<th>Content of the program: content and form</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Participants and leaders introduce themselves</td>
</tr>
<tr>
<td></td>
<td>Educational presentation about current medical approaches</td>
</tr>
<tr>
<td></td>
<td>Educational presentation about self-management</td>
</tr>
<tr>
<td></td>
<td>Discussing relevant individual goals</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Day 2</th>
<th>Educational presentation in one of the topics* with exchange of individual experiences</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Relaxation exercise**</td>
</tr>
<tr>
<td></td>
<td>Selecting individual goals and constructing action plans to achieve these goals</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Day 3-8</th>
<th>Educational presentation in one of the topics* by the leaders with exchange of individual experiences</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Relaxation exercise**</td>
</tr>
<tr>
<td></td>
<td>Participants present and evaluate their individual goals and action plans with feedback/problem solving</td>
</tr>
<tr>
<td></td>
<td>Constructing action plans</td>
</tr>
</tbody>
</table>

One meeting for relatives

*Topics day 2-8: activity pacing and energy conservation, sleep and relaxation, physical exercise, nutritional approaches, economic self-sufficiency, personal relationships, and available treatments.

All educational presentations were made by healthcare professionals at the ME/CFS-centre, Oslo University Hospital.

** Different kind of relaxation exercises were presented during the program.
Intervention group: Lost to follow-up after accepted participation (n = 12) due to ill-health (1), moving to another town (1), and refused the evaluations (10)
Control group: Lost to follow-up after accepted participation (n = 7) due to ill-health (1) and refused the evaluations (6)

**Figure 1.** Flow diagram
Table 2. Characteristics at baseline: mean (SD) or N (%)

<table>
<thead>
<tr>
<th></th>
<th>Intervention (N = 71)</th>
<th>Control (N = 66)</th>
<th>P-values</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>44.0 (11.8)</td>
<td>43.8 (11.6)</td>
<td>0.92</td>
</tr>
<tr>
<td>Diagnose (years)</td>
<td>Median: 3 years</td>
<td>Median: 3 years</td>
<td>0.69&lt;sup&gt;a&lt;/sup&gt;</td>
</tr>
<tr>
<td>Female gender</td>
<td>67 (94.4)</td>
<td>54 (81.8)</td>
<td>0.022&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>Living alone</td>
<td>19 (26.8)</td>
<td>14 (21.2)</td>
<td>0.45&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>Children (number)</td>
<td>1.3 (1.2)</td>
<td>1.4 (1.2)</td>
<td>0.39</td>
</tr>
<tr>
<td>Has children under 18</td>
<td>30 (42.3)</td>
<td>30 (45.5)</td>
<td>0.71&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>Education &gt; 12 years</td>
<td>38 (53.5)</td>
<td>31 (47.0)</td>
<td>0.44&lt;sup&gt;a&lt;/sup&gt;</td>
</tr>
<tr>
<td>Working&lt;sup&gt;c,d&lt;/sup&gt;</td>
<td>3 (4.3)</td>
<td>8 (12.3)</td>
<td>0.09&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>Student</td>
<td>2 (2.8)</td>
<td>0 (0.0)</td>
<td>0.17&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>Energy Envelope Theory&lt;sup&gt;e&lt;/sup&gt;:</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>score ≤ 100</td>
<td>40 (62.5)</td>
<td>45 (69.2)</td>
<td>0.42&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>score ≤ 150</td>
<td>54 (84.4)</td>
<td>52 (80.0)</td>
<td>0.52&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>SF-36 – physical functioning</td>
<td>45.8 (18.2)</td>
<td>46.2 (20.2)</td>
<td>0.91</td>
</tr>
<tr>
<td>Fatigue Severity Scale</td>
<td>56.6 (5.6)</td>
<td>58.0 (4.5)</td>
<td>0.11</td>
</tr>
<tr>
<td>Self-efficacy</td>
<td>16.3 (3.0)</td>
<td>16.6 (2.9)</td>
<td>0.58</td>
</tr>
<tr>
<td>Illness Cognition Questionnaire – acceptance</td>
<td>13.1 (3.4)</td>
<td>13.8 (2.9)</td>
<td>0.25</td>
</tr>
<tr>
<td>SF-36 – physical component summary&lt;sup&gt;e&lt;/sup&gt;</td>
<td>24.2 (6.8)</td>
<td>24.1 (6.8)</td>
<td>0.91</td>
</tr>
<tr>
<td>SF-36 – mental component summary&lt;sup&gt;e&lt;/sup&gt;</td>
<td>37.5 (9.5)</td>
<td>38.9 (11.1)</td>
<td>0.43</td>
</tr>
</tbody>
</table>

All analyzed by independent-samples t-test except:

<sup>a</sup> Analyzed by Mann-Whitney U-test
<sup>b</sup> Analyzed by chi-squared test
<sup>c</sup>1 missing in intervention group
<sup>d</sup>1 missing in control group
<sup>e</sup>N = 64 in intervention group and N = 65 in control group
Table 3a. Efficacy variables at baseline and six-month follow-up

<table>
<thead>
<tr>
<th></th>
<th>Intervention (N = 63)</th>
<th>Control (N = 62)</th>
<th>Results from mixed model analysisa</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Baseline Mean (SD)</td>
<td>Six-month follow-up Mean (SD)</td>
<td>Change Mean (95 % CI)</td>
</tr>
<tr>
<td>SF-36 – physical functioningb</td>
<td>47.0 (18.3)</td>
<td>47.5 (21.2)</td>
<td>0.6 (-2.9, 4.0)</td>
</tr>
<tr>
<td>Fatigue severity scale</td>
<td>56.3 (5.8)</td>
<td>56.0 (6.8)</td>
<td>-0.2 (-1.7, 1.3)</td>
</tr>
<tr>
<td>Self-efficacyb</td>
<td>16.3 (2.8)</td>
<td>16.7 (3.1)</td>
<td>0.4 (-0.4, 1.1)</td>
</tr>
<tr>
<td>Illness cognition questionnaire – acceptanceb</td>
<td>13.3 (3.5)</td>
<td>14.3 (3.6)</td>
<td>0.9 (0.3, 1.6)</td>
</tr>
<tr>
<td>SF-36 – physical component summaryc</td>
<td>24.6 (7.0)</td>
<td>24.8 (7.3)</td>
<td>0.2 (-1.4, 1.7)</td>
</tr>
<tr>
<td>SF-36 – mental component summaryc</td>
<td>37.3 (10.0)</td>
<td>37.4 (11.2)</td>
<td>0.1 (-2.5, 2.7)</td>
</tr>
</tbody>
</table>

aComparison of intervention and control group.

b1 missing in control group

cN = 61 in both intervention and control group
<table>
<thead>
<tr>
<th></th>
<th>Intervention (N = 59)</th>
<th>Control (N = 59)</th>
<th>Results from mixed model analysis&lt;sup&gt;a&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Baseline Mean (SD)</td>
<td>One-year follow-up Mean (SD)</td>
<td>P-values</td>
</tr>
<tr>
<td>SF-36 – physical functioning</td>
<td>48.1 (17.7)</td>
<td>48.9 (20.9)</td>
<td>0.76</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0.8 (4.2, 5.7)</td>
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<td></td>
<td></td>
<td>46.5 (21.0)</td>
<td>-0.3 (-5.4, 4.9)</td>
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<tr>
<td></td>
<td></td>
<td>0.76</td>
<td></td>
</tr>
<tr>
<td>Fatigue severity scale</td>
<td>56.0 (5.9)</td>
<td>56.4 (6.9)</td>
<td>0.66</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0.4 (-1.4, 2.2)</td>
<td></td>
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<td></td>
<td></td>
<td>58.5 (3.9)</td>
<td>-1.4 (-3.0, 0.1)</td>
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<td></td>
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<td>0.66</td>
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<td></td>
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<td>57.1 (6.7)</td>
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<td></td>
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<td>0.12</td>
<td>0.55</td>
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<tr>
<td>Self-efficacy</td>
<td>16.5 (2.8)</td>
<td>16.3 (3.4)</td>
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<td></td>
<td></td>
<td>-0.2 (-1.1, 0.7)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>16.7 (2.9)</td>
<td>-0.5 (-1.2, 0.1)</td>
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<td></td>
<td>0.11</td>
<td>0.55</td>
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<tr>
<td></td>
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<td>16.2 (3.2)</td>
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</tr>
<tr>
<td></td>
<td></td>
<td>0.11</td>
<td></td>
</tr>
<tr>
<td>Illness cognition questionnaire – acceptance</td>
<td>13.3 (3.5)</td>
<td>14.0 (3.5)</td>
<td>0.7</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0.7 (0.1, 1.4)</td>
<td>0.030</td>
</tr>
<tr>
<td></td>
<td></td>
<td>14.0 (3.0)</td>
<td>14.5 (3.1)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0.5 (-0.1, 1.1)</td>
<td>0.12</td>
</tr>
<tr>
<td>SF-36 – physical component summary&lt;sup&gt;b&lt;/sup&gt;</td>
<td>24.9 (7.0)</td>
<td>24.7 (8.0)</td>
<td>0.83</td>
</tr>
<tr>
<td></td>
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<td>-0.2 (-2.3, 1.8)</td>
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<tr>
<td></td>
<td></td>
<td>24.0 (7.0)</td>
<td>24.2 (8.5)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0.1 (-2.0, 2.2)</td>
<td>0.91</td>
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<td></td>
<td></td>
<td>0.91</td>
<td>0.94</td>
</tr>
<tr>
<td>SF-36 – mental component summary&lt;sup&gt;b&lt;/sup&gt;</td>
<td>37.7 (9.9)</td>
<td>39.1 (10.6)</td>
<td>1.4</td>
</tr>
<tr>
<td></td>
<td></td>
<td>1.4 (-1.0, 3.8)</td>
<td>0.26</td>
</tr>
<tr>
<td></td>
<td></td>
<td>39.3 (10.9)</td>
<td>40.5 (8.8)</td>
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<td></td>
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<td>1.2 (-1.2, 3.5)</td>
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<td></td>
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<td>0.32</td>
<td>0.88</td>
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</table>

<sup>a</sup>Comparison of intervention and control group
<sup>b</sup>1 missing in intervention group