ARTICLE CLASS OF EVIDENCE

# Self-management program improves participation in patients with neuromuscular disease

## A randomized controlled trial

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## Abstract

### Objective

To investigate the effectiveness of Energetic, a self-management group program combining aerobic training, energy conservation management, and relapse prevention to improve social participation in patients with neuromuscular disease (NMD) and chronic fatigue.

### Methods

In this multicenter, assessor-blinded, 2-armed randomized controlled trial with repeated measurements, 53 patients with various types of NMD and chronic fatigue were randomly allocated to Energetic, a 4-month group intervention, or to usual care. The primary endpoint was social participation assessed with the Canadian Occupational Performance Measure (COPM) performance scale immediately postintervention. Secondary outcomes included COPM satisfaction scale, 6-Minute Walk Test (6MWT), and Checklist Individual Strength–subscale fatigue. Participants were followed for 11 months postintervention. Data were analyzed with linear models that account for repeated measurements.

### Results

Directly after intervention, the mean group difference for COPM-performance was 1.7 (95% confidence interval [CI] 1.0–2.4; $p < 0.0001$) in favor of the intervention group (n = 29), adjusted for baseline, sex, diagnosis, and work status. This effect was retained at 11 months follow-up (0.9; 95% CI 0.0–1.7; $p = 0.049$). The COPM satisfaction scale and 6MWT improved more in the intervention group compared to usual care. After 3 and 11 months follow-up, most beneficial effects on social participation and functional endurance were retained.

### Conclusion

Energetic led to sustainable improvements in social participation and functional endurance compared to usual care in patients with NMD and chronic fatigue.

### Clinicaltrials.gov identifier

NCT02208687.

### Classification of evidence

This study provides Class III evidence that a combination of aerobic training, energy conservation management, and relapse prevention improves social participation in patients with NMD and chronic fatigue.

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## Glossary

**6MWT** = 6-Minute Walk Test; **ACS** = Activity Card Sort; **AET** = aerobic exercise training; **ARR** = absolute risk reduction; **CBT** = cognitive behavioral therapy; **CI** = confidence interval; **CIS** = Checklist Individual Strength; **COPM** = Canadian Occupational Performance Measure; **ECM** = energy conservation management; **FSHD** = facioscapulohumeral dystrophy; **GSES** = General Self-Efficacy Scale; **HADS** = Hospital Anxiety and Depression Scale; **IBM** = inclusion body myositis; **MM** = mitochondrial myopathies; **NMD** = neuromuscular disease; **NNT** = number needed to treat; **RUMC** = Radboud University Medical Center.

More than 60% of patients with a neuromuscular disease (NMD) report chronic fatigue as their most disabling symptom leading to reduced social participation.[1,2] As there is no cure for most NMD, interventions aim to influence fatigue-related factors to increase participation.[3] Research has demonstrated that aerobic exercise training (AET) alleviates chronic fatigue in facioscapulohumeral dystrophy (FSHD)[4] and mitochondrial myopathies (MM).[5,6] Cognitive behavioral therapy (CBT) has also proven to diminish chronic fatigue in FSHD[4] and to improve participation in myotonic dystrophy type 1.[7] In multiple sclerosis, several studies investigated energy conservation management (ECM), which aims to support fatigue self-management in daily life to enable participation in valuable occupations.[8,9] However, there are no studies of ECM in NMD.

A recent meta-analysis[10] of 15 studies and 1,696 patients with common disabling disorders has shown that diagnosis explains only 11% of fatigue severity, which increases to 55% when transdiagnostic factors are added such as sex, motivation, and activity level. This notion provides support for a transdiagnostic fatigue-management approach focusing on individual patient characteristics rather than disease characteristics.[10] In this perspective, we developed and implemented a multidisciplinary group program, called Energetic, aimed to optimize social participation and functional endurance in patients with various types of NMD and chronic fatigue. The program combines AET and education about AET with ECM and relapse prevention.[11] Here, we report a pragmatic study on the effectiveness of Energetic compared to usual care in patients with various types of NMD who are chronically fatigued.

## Methods

### Study objectives

The primary objective of this study was to evaluate the effect of Energetic, a 16-week group intervention combining AET, ECM, and relapse prevention on social participation in patients with NMD with chronic fatigue compared to usual care (classification of evidence: Class III). The secondary objective was to evaluate the effects of Energetic on fatigue, functional endurance, activity engagement, mood, anxiety, self-efficacy, and perceived caregiver burden (classification of evidence: Class III).

### Study design

We conducted a multicenter, assessor-blinded, 2-armed randomized controlled trial with repeated measurements in the Netherlands during a 15-month study period. The coordination of the study took place at Radboud University Medical Center (RUMC), while the intervention was, besides at RUMC, also offered in 2 other clinical settings: a regional rehabilitation center (Klimmendaal in Arnhem) and a community health center in primary care (Buitenlust in Venray).

### Participants

Patients were recruited at the Departments of Rehabilitation, Neurology, and Internal Medicine of the RUMC as well as in the other participating centers. The Dutch patient association for NMD (Spierziekten Nederland) facilitated recruitment by posting study information (website, magazine, and mailing). Referred or otherwise interested patients were provided with written study information and contacted by telephone by the primary investigator (Y.V.) to answer any remaining questions related to study participation. The primary informal caregiver of the patient was invited to coparticipate. When patients expressed their willingness to participate and screening of the inclusion and exclusion criteria indicated potential eligibility, outpatient clinic appointments with a rehabilitation physician and occupational therapist at RUMC were planned for definitive inclusion and obtaining signed informed consent. The inclusion criteria were (1) age 18 years or older, (2) diagnosis of NMD determined by a neurologist using established criteria, and (3) subjective experience of chronic fatigue with a clear effect on daily life and social participation determined by an occupational therapist. In addition, the occupational therapist assessed the motivation and readiness to change as well as the ability to formulate at least 3 personalized participation goals. This study purposely included patients with various NMD, but the recruitment was preferentially focused on patients with FSHD, MM, and inclusion body myositis (IBM), as RUMC hosts (inter)nationally recognized expert centers for these specific NMDs. The exclusion criteria were (1) major cardiorespiratory problems that precluded participation in AET, (2) pregnancy, (3) limited life expectancy (<5 years) due to known comorbid conditions, and (4) having participated in the Energetic program or a similar intervention before. Before definitive inclusion, all participants were assessed by a psychologist for possible depressive symptoms and other psychiatric or cognitive symptoms, including addiction

problems, to determine whether patients could participate in the Energetic program.[11]

## Randomization and masking

Enrolled patients were randomized in a 1:1 ratio to either an intervention group receiving the Energetic program or a control group receiving usual care. Randomization, performed by an independent statistician (H.G.), was based on a computerized minimization algorithm with the following minimization factors: sex (men vs women), work status (work vs no work), diagnosis (FSHD, IBM, MM vs other NMD). All outcomes (of both patients and caregivers) were assessed by blinded and independent (occupational therapy) research assistants and subsequently entered into a digital and validated database (Castor). Patients, caregivers, and therapists could not be blinded, but all participants were urged not to discuss their allocation status with the assessors. At all follow-up assessments, assessors recorded whether their blinding might have been broken.

## Standard protocol approvals, registrations, and patient consents

Full ethical approval was granted by the medical ethical committee of the region Arnhem-Nijmegen (NL47624.091.14) and the executive boards of all participating centers. All patients provided oral and written informed consent. The trial was registered at clinicaltrials.gov (NCT02208687). Details of the study protocol were published previously.[11]

## Intervention

Within 2 weeks after randomization, the intervention group started the Energetic program, which was offered to small subgroups with a minimum of 4 and a maximum of 8 patients. Although a group program was applied, AET and self-management strategies were individualized as much as possible. The program was delivered by a physical and occupational therapist and consisted of the following 4 modules, which have been described in detail previously:[11]

### Aerobic exercise training

During 16 weeks, patients received individually tailored AET twice a week during the first 9 weeks (part 1) and once a week during the last 7 weeks (part 2) with guidance from the physical therapist. Patients were expected to perform physical exercises at home once (part 1) and twice (part 2) weekly, so that that the overall AET load amounted to 3 sessions of 30 minutes per week during the entire intervention period of 4 months. Training intensity was aimed at 50%–70% of the maximum heart rate, based on a maximal cycling exercise test. Training intensity was fine-tuned individually based on the heart rate recovery guided by a heart rate monitor that patients privately purchased or borrowed from the physical therapist.[12] Patients used the same heart rate monitor at home. They reported the results of their home exercises during the next group session in the clinical setting. The training included different exercises, such as walking on a treadmill, cycling on a home trainer, rowing on an ergometer, and using a cross-trainer. The type of exercise depended on personal preference, motor abilities, and practical possibilities.

### Exercise education

In three 60-minute sessions during the first 3 weeks of the intervention period, patients were educated about general physical and aerobic exercise training principles in relation to NMD. These sessions were given by a physical therapist.

### Energy conservation management

During eight 90-minute sessions spread across the intervention period, ECM training was supported by an occupational therapist. These sessions included education and discussion, extended by individual goal-setting, practicing activities, and performing homework activities with the aim to learn and apply energy conservation strategies in daily life.[8,13]

### Implementation and relapse prevention

During 10 group sessions, physical and occupational therapists or sports trainers supported and empowered the patients with the implementation of AET and ECM in daily life, with a specific focus on finding a sustainable way to exercise at home. Caregivers were involved in 2 of these sessions. In this module, different types of sports and exercises were explored that would be suitable to continue in the home environment.

In addition, a booster session of 2 hours with the physical and occupational therapists was organized 2 months after the end of the intervention period to reinforce previously learned strategies and skills.

Patients in the control group were not prescribed (or withheld) any specific intervention, which meant that some received physical therapy in primary care, other forms of multidisciplinary rehabilitation care, or no intervention at all.[14] Patients in both the intervention and control groups were not discouraged from taking part in any additional activities during the study period.

Individual therapy compliance was recorded by the therapists regarding attendance of both the training and education sessions. When applicable, patients were asked about their reasons for noncompliance. At every assessment in both the intervention and control groups, the consultation of health care and social support professionals was recorded with a questionnaire focused on the preceding months.

## Procedure

The Energetic program was administered by a single occupational therapist and a single physical therapist in each of the 3 health care settings. All therapists had received a 1-day education program about the pathophysiology and functional consequences of different NMDs, the Energetic study protocol, and the content of the Energetic program. During the study, all therapists joined 3 additional intervision sessions to

discuss their experiences in order to support uniformity in the delivery of the program. Irregularities affecting protocol adherence were registered by the primary researcher (Y.V.).

Outcome data for patients and caregivers were collected before randomization (T0), immediately after the 4-month intervention period (T1), at 3 months follow-up (T2), and at 11 months follow-up (T3). At baseline, demographic and clinical characteristics (age, sex, education level, work status, type of NMD) were collected for patients and their caregivers.

## Outcome measures

The primary endpoint was the mean score of patient's self-rated performance of 3 to 5 self-selected daily occupations identified with the Canadian Occupational Performance Measure (COPM) performance scale at T1.[15] Through a semi-structured interview, patients identified and prioritized problems in their occupational performance. These occupations were subsequently rated on a 10-point scale for perceived performance (1 = not able at all; 10 = perfectly able). Previous studies have shown that the COPM is a valid and responsive instrument to assess participation problems experienced by patients in self-care, productivity, and leisure.[16,17]

We selected COPM-performance as the primary outcome, and used Checklist Individual Strength–fatigue subscale (CIS-fatigue) as a secondary outcome, because the main goal of our intervention was to improve social participation based on behavioral change rather than alter dysfunctional cognitions or experienced fatigue. There are no objective measures of social participation that would be able to pick up subtle, but important changes regarding family life, leisure, work, and maintaining relationships. Therefore, a personalized goal attainment tool seemed to be the most suitable outcome measure.[18–20]

Several secondary outcomes were selected to explore and better understand any underlying physical, psychological, and behavioral changes associated with improved social participation. These included patient's self-rated satisfaction with their performance assessed with the COPM satisfaction scale, experienced fatigue assessed with the CIS-fatigue,[21] functional endurance assessed with the 6-Minute Walk Test (6MWT),[22,23] activity engagement assessed with the Activity Card Sort (ACS),[24,25] mood and anxiety assessed with the Hospital Anxiety and Depression Scale (HADS),[26] and self-efficacy assessed with the General Self-Efficacy Scale (GSES).[27] In addition, caregivers were assessed for perceived caregiver burden with the Zarit Burden Interview.[28] Blinded assessors monitored adverse events at each assessment.

## Power

Based on a pilot study of 13 patients using COPM-performance as a primary outcome,[29] an estimate of the improvement with Energetic compared to usual care was set at 2.0 and the SD of change in each group at 1.7. Based on these values, 13 participants would be needed in each group to obtain a power of 80% using a 2-sided $t$ test ($\alpha$ level 5%) in order to detect a group difference in COPM-performance at T1. Taking into account a dropout rate of 10%, a total of 30 participants would be required. However, to meet the required estimates for determining the cost-effectiveness (cost analyses reported separately), our aim was to include at least 50 patients.

## Statistical analysis

We used generalized estimating equations in a linear regression model to study the intervention effects for all primary and secondary outcomes. The primary hypothesis was tested solely based on the COPM-performance. The value of the dependent variable at baseline (T0) as well as treatment (Energetic vs control), time (T1, T2, T3), treatment × time interaction, sex, diagnosis, and work status were included as independent variables in all analyses. To account for varying dependencies due to the unequal time periods between the measurements, no prior assumptions were made on the within-individual dependencies between the measurements over time (expressed in the covariance matrix of the models). Clinical setting was assigned randomly (as it was assigned to the location that was closest to the patient's home). The variable was not added to the model as an independent variable, because only a few patients were treated in a primary health care setting. Estimates of the regression measures were calculated, including their 95% confidence intervals (CIs), based on the robust sandwich estimates of the standard errors. All statistical analyses were based on the intention-to-treat principle. To evaluate the effects of missing variables on the observed results, a sensitivity analysis using a last observation carried forward imputation technique was done for the primary outcome.

In a post hoc analysis, we used the Fisher exact test to compare the proportion of patients in each group who reached the minimal clinically important change on COPM-performance from baseline. The threshold for this minimal clinically important change was defined as a difference of at least 2 points.[30] In addition, the number needed to treat (NNT) was calculated as the reciprocal of the absolute risk reduction (ARR). All statistical analyses were done by an independent statistician (M.A.J.) using Rstudio and SPSS version 22.

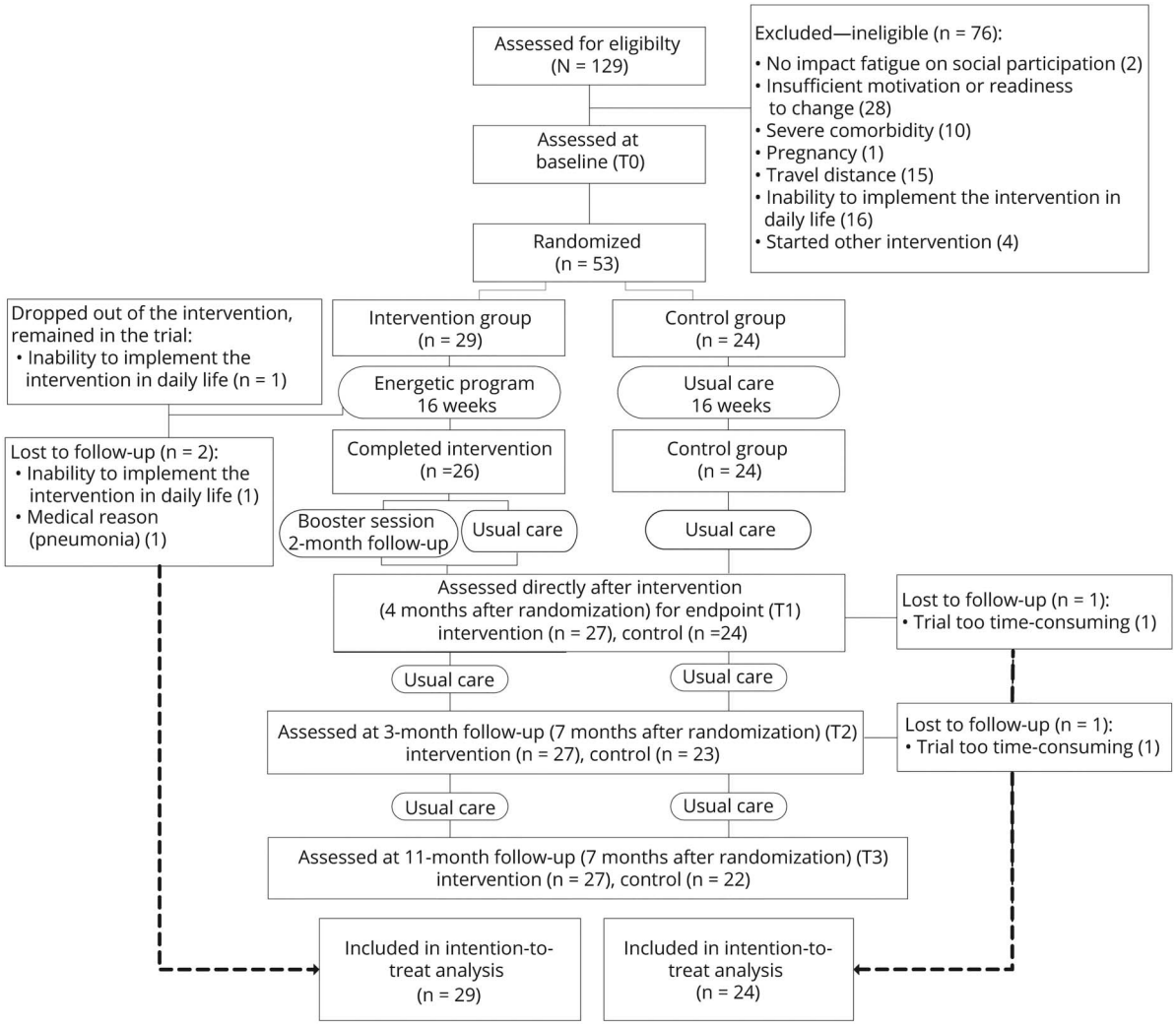
## Data availability

Related study protocols and anonymized data will be shared with qualified investigators on request, including stated data sharing conditions (research question, ownership, security/protection, and confidentiality of data) until 2026.

# Results

The flowchart of the study participants is shown in figure 1. Of the 129 patients who were assessed for eligibility, 53 met the inclusion criteria and were enrolled from July 22, 2014, to

**Figure 1** Flowchart of the study participants




September 1, 2015. Twenty-two of the 29 patients in the intervention group (76%) and 19 of the 24 patients in the control group (79%) had a caregiver who was able and willing to co-participate. Patients in the intervention group received Energetic in 3 different settings: (1) 3 groups in a university hospital (n = 13), 3 groups in a rehabilitation center (n = 12), and 1 group in a community health center (n = 4). The reasons for nonparticipation by patients were (1) insufficient motivation or readiness to change, (2) inability to implement the intervention in their personal agenda, and (3) travel distance. During the intervention period, 3 patients withdrew from the intervention group, of whom 2 were lost to follow-up. During the follow-up period, 2 patients withdrew from the control group and were lost to follow-up. In addition, 8 caregivers were lost to follow-up during the entire study period of 15 months (intervention group n = 5; control group n = 3).

In table 1, the baseline characteristics of the patients and caregivers are presented for both the intervention and control groups. The groups were similar at baseline with respect to age, sex, work status, education level, and baseline values of the primary and secondary outcomes. Type of NMD was also similarly distributed among both groups. Most caregivers were partners of the patients (n = 36); others were siblings (n = 3) or children (n = 2).

Directly after the 4-month treatment period (T1), the intervention group showed significantly higher mean COPM-performance scores compared to the control group adjusted for baseline, sex, diagnosis, and work status. This effect was still significant at 11 months follow-up and after correction for possible confounders. The adjusted mean group differences in COPM-performance were 1.7 (95% CI 1.0–2.4) at T1, 0.79 (95% CI −0.2–1.8) at T2, and 0.85 (95% CI 0.0–1.7) at T3 (table 2, figure 2). Sensitivity analyses of the missing values for COPM-performance scores (n = 2) showed a slightly lower mean group difference at T1 (1.5; 95% CI 0.9–2.2) compared to the primary analysis. Post hoc analysis showed that the proportion of patients who had attained a clinically relevant

**Table 1** Characteristics of the participants

| | Intervention group (n = 29) | Control group (n = 24) |
|---|---|---|
| **Patients** | | |
| **Age, y** | 52 (37–63) | 50 (41–60) |
| **Sex, male** | 8/29 (28) | 9/24 (38) |
| **Education level**[a] | | |
| **High** | 11/28 (39) | 4/23 (17) |
| **Middle** | 11/28 (39) | 16/23 (70) |
| **Low** | 6/28 (21) | 3/23 (13) |
| **Paid employment** | 13/29 (45) | 12/24 (50) |
| **Type of NMD** | | |
| **Facioscapulohumeral dystrophy** | 4/29 (14) | 1/24 (4) |
| **Inclusion body myositis** | 3/29 (10) | 2/24 (8) |
| **Mitochondrial myopathy** | 8/29 (24) | 10/24 (42) |
| **Hereditary motor and sensory neuropathy** | 3/29 (10) | 2/24 (8) |
| **Myasthenia gravis** | 2/29 (7) | 2/24 (8) |
| **Myotonic dystrophy type 1** | 2/29 (7) | 1/24 (4) |
| **Hereditary spastic paraplegia** | 1/29 (3) | 1/24 (4) |
| **Chronic idiopathic axonal polyneuropathy** | — | 1/24 (4) |
| **Congenital myopathy** | — | 1/24 (4) |
| **McArdle syndrome** | — | 1/24 (4) |
| **Congenital fiber type disproportion** | 1/29 (3) | 1/24 (4) |
| **Limb girdle muscular dystrophy** | — | 1/24 (4) |
| **Autosomal dominant distal and anterior dystrophy** | — | 1/24 (4) |
| **Kennedy syndrome** | 1/29 (3) | — |
| **Hyperkalemic periodic paralysis** | 1/29 (3) | — |
| **Duchenne carrier** | 2/29 (7) | — |
| **Postpolio syndrome** | 1/29 (3) | — |
| **Time since diagnosis, y** | 7 (0–41) | 2 (0–39) |
| **Caregivers** | | |
| **Partners** | 19 (86) | 15 (88) |
| **Age, y** | 55 (45–65) | 53 (47–58) |
| **Sex, male** | 13/22 (59) | 12/18 (67) |
| **Educational level** | | |
| **High** | 9/22 (41) | 2/18 (12) |
| **Middle** | 13/22 (59) | 14/18 (82) |
| **Low** | — | 1/18 (6) |
| **Paid employment** | 16/22 (73) | 15/18 (83) |

Abbreviation: NMD = neuromuscular disease.
Data are median (interquartile range) or n/N (%). Some percentages do not sum up to 100% due to rounding.
[a] Data missing from one patient in the intervention group and one in the control group.

**Table 2** Observed means (SDs) and estimated mean group differences (95% confidence intervals [CIs]) (adjusted for baseline values, sex, work status, and diagnosis) for all outcome measures by point of measurement (patients and caregivers)

| | Baseline (T0) | | Immediately post intervention (T1) | | 3-Month follow-up (T2) | | 11-Month follow-up (T3) | | Intervention effect at T1 | | Intervention effect at T2 | | Intervention effect at T3 | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | N | Mean (SD) | N | Mean (SD) | N | Mean (SD) | N | Mean (SD) | Mean (95% CI) | *p* Value | Mean (95% CI) | *p* Value | Mean (95% CI) | *p* Value |
| **Canadian Occupational Performance Measure-performance scale (COPM-P score 1–10), higher scores indicate better self-perceived performance of prioritized occupations** | | | | | | | | | | | | | | |
| **Intervention** | 29 | 3.8 (1.3) | 27 | 6.5 (1.4) | 27 | 5.9 (2.0) | 27 | 5.7 (1.9) | 1.7 (1.0, 2.4)[a] | < 0.0001 | 0.8 (−0.2, 1.8) | 0.11 | 0.9 (0.0, 1.7) | 0.049 |
| **Control** | 24 | 3.7 (1.2) | 24 | 4.7 (1.4) | 23 | 5.1 (1.9) | 22 | 4.8 (1.8) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Canadian Occupational Performance measure-satisfaction scale (COPM-S score 1–10), higher scores indicate better self-perceived satisfaction with performance of prioritized occupations** | | | | | | | | | | | | | | |
| **Intervention** | 29 | 3.2 (1.4) | 27 | 6.2 (1.6) | 27 | 6.0 (1.8) | 27 | 5.5 (1.5) | 2.1 (1.2, 3.0) | <0.0001 | 1.3 (0.3, 2.3) | <0.0001 | 0.7 (−0.3, 1.7) | 0.14 |
| **Control** | 24 | 3.1 (1.5) | 24 | 4.1 (1.7) | 23 | 4.7 (1.9) | 22 | 4.8 (1.6) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **6-Minute Walk Test (meters), higher scores indicate more meters walked in 6 minutes** | | | | | | | | | | | | | | |
| **Intervention** | 28 | 438 (95) | 27 | 479 (97) | 27 | 474 (94) | 25 | 474 (95) | 30.3 (12.4, 48.2) | 0.00073 | 38.5 (11.3, 65.7) | 0.0046 | 40.4 (16.0, 64.8) | 0.00092 |
| **Control** | 24 | 445 (95) | 23 | 463 (94) | 21 | 450 (93) | 20 | 440 (89) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Checklist Individual Strength–subscale fatigue (score 0–56), lower scores indicate lower self-perceived impact of fatigue[b]** | | | | | | | | | | | | | | |
| **Intervention** | 28 | 41.1 (1.0) | 26 | 31.0 (5.5) | 27 | 33.0 (11.9) | 26 | 36.8 (10.4) | −2.2 (−4.7, 0.3) | 0.081 | −3.9 (−8.5, 0.8) | 0.095 | −1.8 (−7.5, 3.9) | 0.53 |
| **Control** | 24 | 44.9 (8.9) | 22 | 35.0 (4.9) | 21 | 40.0 (9.3) | 18 | 40.4 (12.2) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Activity Card Sort (%), higher percentages indicate more participation in different activities** | | | | | | | | | | | | | | |
| **Intervention** | 29 | 72.1 (12.0) | 27 | 65.6 (10.4) | 27 | 75.5 (12.4) | 26 | 74.1 (13.5) | 4.3 (−0.02, 8.5) | 0.047 | 4.8 (−1.4, 11.0) | 0.12 | −1.1 (−7.7, 5.6) | 0.75 |

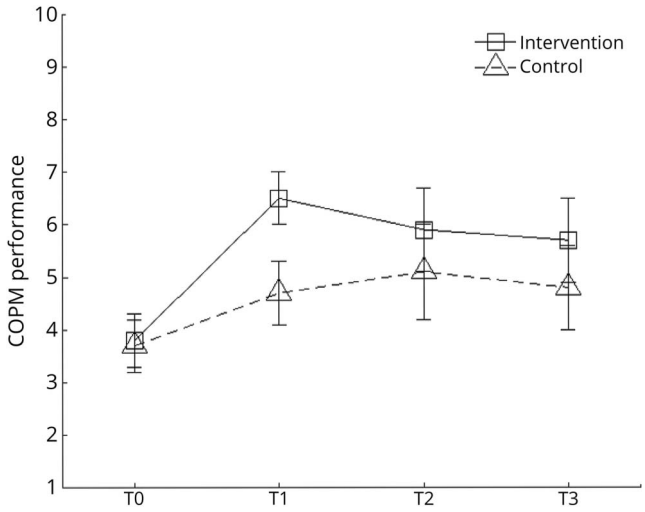
Continued

**Table 2** Observed means (SDs) and estimated mean group differences (95% confidence intervals [CIs]) (adjusted for baseline values, sex, work status, and diagnosis) for all outcome measures by point of measurement (patients and caregivers) *(continued)*

| | Baseline (T0) | | Immediately post intervention (T1) | | 3-Month follow-up (T2) | | 11-Month follow-up (T3) | | Intervention effect at T1 | | Intervention effect at T2 | | Intervention effect at T3 | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | N | Mean (SD) | N | Mean (SD) | N | Mean (SD) | N | Mean (SD) | Mean (95% CI) | *p* Value | Mean (95% CI) | *p* Value | Mean (95% CI) | *p* Value |
| **Control** | 24 | 76.5 (15.3) | 23 | 65.2 (12.6) | 21 | 75.0 (17.5) | 21 | 79.2 (17.0) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **General Self-Efficacy Scale (10–40), higher scores indicate better self-efficacy** | | | | | | | | | | | | | | |
| **Intervention** | 28 | 34.1 (4.7) | 26 | 32.2 (3.3) | 27 | 31.3 (4.0) | 26 | 32.1 (3.3) | 1.0 (−0.80, 2.8) | 0.27 | 0.7 (−1.2, 2.5) | 0.46 | −0.6 (−2.3, 1.1) | 0.47 |
| **Control** | 24 | 33.8 (4.5) | 22 | 31.2 (4.2) | 21 | 30.6 (3.3) | 17 | 32.9 (4.5) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Hospital Anxiety and Depression Scale–anxiety (0–21), lower scores indicate lower levels of anxiety**[b] | | | | | | | | | | | | | | |
| **Intervention** | 28 | 6.0 (3.2) | 26 | 5.0 (3.2) | 27 | 4.3 (2.6) | 26 | 4.5 (2.4) | −0.4 (−1.6, 0.73) | 0.46 | −1.2 (−2.8, 0.41) | 0.14 | −1.0 (−2.3, 0.37) | 0.15 |
| **Control** | 24 | 5.3 (3.2) | 22 | 5.0 (3.0) | 21 | 5.1 (3.0) | 17 | 5.3 (3.6) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Hospital Anxiety and Depression Scale–depression (0–21), lower scores indicate lower levels of depressed mood**[b] | | | | | | | | | | | | | | |
| **Intervention** | 28 | 5.4 (3.5) | 26 | 4.5 (3.0) | 27 | 4.7 (3.1) | 26 | 4.3 (2.1) | −2.1 (−3.4, −0.71) | 0.0022 | −1.9 (−3.3, −0.37) | 0.013 | −1.6 (−3.2, 0.056) | 0.053 |
| **Control** | 24 | 4.8 (2.6) | 22 | 5.7 (3.9) | 21 | 5.3 (3.8) | 17 | 4.8 (3.4) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |
| **Zarit Burden Index (score 0–88), lower scores indicate less caregiver burden**[c] | | | | | | | | | | | | | | |
| **Intervention** | 22 | 14.4 (8.1) | 20 | 15.7 (9.2) | 19 | 15.7 (10.3) | 17 | 18.8 (12.0) | 0.9 (−1.96, 3.84) | 0.52 | −1.2 (−4.54,2.06) | 0.45 | 3.7 (−0.12,7.4) | 0.053 |
| **Control** | 19 | 15.3 (9.3) | 17 | 16. 1 (8.0) | 16 | 17.8 (10.2) | 13 | 16.7 (9.7) | 0.0 (ref) | | 0.0 (ref) | | 0.0 (ref) | |

Abbreviation: CI = confidence interval.
Descriptive data are presented as means (SD). For all measures, unless indicated otherwise, an increase in the score over time reflects improvement. Intervention effects are presented as mean group differences (95% CIs). For patients, group differences were estimated using generalized estimating equations in a linear model with adjustments for baseline values, sex, work status, and diagnosis.
[a] Primary outcome.
[b] Decrease in the score over time reflects improvement.
[c] Caregiver outcome.

**Figure 2** Means with 95% confidence intervals of the Canadian Occupational Performance Measure, Performance score (COPM) at different time points




T0: baseline; T1: postintervention (4 months); T2: 3 months follow-up (7 months after randomization); T3: 11 months follow-up (15 months after randomization) for the intervention group and control group.

improvement on COPM-performance (≥2 points) at T1 was larger for the intervention group (21/29; 72%) than for the control group (6/24; 25%) (Fisher exact test $p = 0.001$). Clinically relevant deterioration only occurred in the control group (7/24; 29%) (Fisher exact test $p = 0.001$). As a result, the NNT for Energetic was 2.3 (95% CI 1.5–4.8) with an ARR of 44% (95% CI 21%–68%).

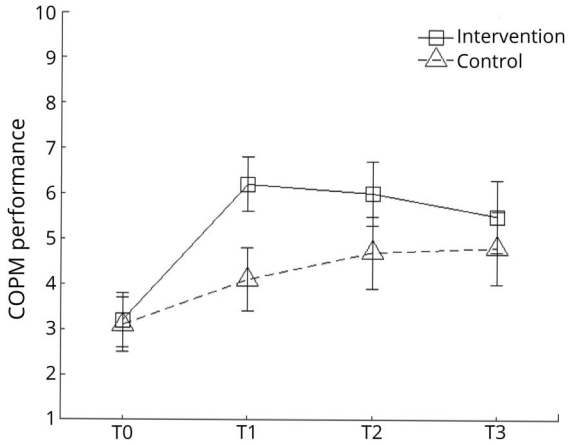
The adjusted mean group differences in COPM-satisfaction score were 2.1 (95% CI 1.2–3.0) at T1, 1.3 (95% CI 0.31–2.3) at T2, and 0.70 (−0.25–1.7) at T3 (table 2, figure 3), indicating significant effects in favor of the Energetic group directly postintervention and at 3 months follow-up. Significant effects in favor of the intervention group were also found on the 6MWT (T1-T3), HADS-Depression (T1 and T2), and ACS (T1) (table 2). There were no significant group differences for CIS-fatigue, GSES, or HADS-anxiety. The perceived caregiver burden was low for both groups and did not show significant changes in time. At baseline, 19 caregivers in the intervention group (79%) and 15 caregivers in the control group (86%) scored lower than 24 points.[31]

Patients in the control group received their usual care consisting of various interventions during the intervention period. Thirteen patients (54%) received multidisciplinary rehabilitation at a rehabilitation center, and 6 patients (25%) received some form of allied health care in the community. Five patients (21%) received no intervention at all. In the intervention group, 13 patients (45%) visited additional allied health care professionals (e.g., physical therapist, occupational therapist, dietician, psychologist, speech-language therapist) alongside Energetic. There were no irregularities affecting protocol adherence by therapists. In 5 patients (17%), noncompliance to one of the sessions was reported by the therapist. With regard to adverse events, one patient (intervention group) was admitted to a hospital for pneumonia and one patient (intervention group) dropped out due to the inability to continue participation in the Energetic program (figure 1). Immediately postintervention, masking of assessors was broken in 9 of the 53 cases (17%). At 3 and 11 months follow-up, no further demasking occurred.

## Discussion

This study showed that the Energetic self-management program resulted in a significant improvement in social participation (COPM-performance) compared to usual care in a heterogeneous group of patients with NMD who were chronically fatigued. A post hoc analysis showed that a significantly larger proportion of patients in the intervention group (72%) achieved a clinically relevant improvement of ≥2.0 points on the COPM-performance compared to the control group (25%) with an NNT of 2.3 for the intervention group.[30] In addition, patients' satisfaction with their performance (COPM-satisfaction scale) and functional endurance, measured with the 6MWT, improved more after the Energetic program compared to usual care, even though walking was not the preferred type of exercise for many patients. After 3 and 11 months follow-up, most beneficial effects on social participation and functional endurance were retained, although the effects on social participation tended to become smaller over time. Remarkably, experienced fatigue (CIS-fatigue) and self-efficacy (GSES) did not significantly respond

**Figure 3** Means with 95% confidence intervals of the Canadian Occupational Performance Measure, Satisfaction score (COPM) at different time points




T0: baseline; T1: postintervention (4 months); T2: 3 months follow-up (7 months after randomization); T3: 11 months follow-up (15 months after randomization) for the intervention group and control group.

to treatment. This pattern of results may suggest that the observed improvement in social participation was mediated by changes in physical endurance and activity engagement rather than by changes in experienced fatigue or self-efficacy.

Contrary to our expectations and to the results of our pilot study,[29] the Energetic program did not show a significant effect on experienced fatigue (CIS-fatigue) compared to usual care. Both the control and intervention group reported less experienced fatigue after the intervention period. A possible explanation may be a lack of contrast between both groups, as 19 patients in the control group received some form of rehabilitation treatment or primary care physical therapy. Another explanation may be that Energetic primarily focused on optimizing daily activities and social participation through ECM and implementing behavioral change in daily life, rather than on altering perceptions and cognitions concerning chronic fatigue, which is the focus of CBT.[10,13,32] Notably, optimizing daily activities did not imply performing more activities, which was indicated by a decrease in the ACS score in both groups immediately post intervention, albeit smaller in the Energetic group. Apparently, patients needed to initially reduce the number of activities to eventually be able to optimize their activity pattern.[33]

We expected that the self-efficacy of patients would change over time as a result of training self-management and problem-solving skills. However, patients in the control and intervention group already had high general self-efficacy scores at baseline and showed minimal changes over time.[34] With regard to emotional functioning, patients' mood (HADS-depression) appeared to be more responsive to the Energetic program than anxiety (HADS-anxiety), as depressive symptoms tended to get worse in the control group, whereas they improved in the Energetic group. Caregiver burden proved to be too low to pick up any treatment effects.

Unlike previous research in NMD, our study investigated an already existing multidisciplinary group intervention in chronically fatigued patients with various types of NMD, now implemented in different clinical settings based on a pragmatic study goal and design.[35] We combined (self-management education about) AET, ECM, and support for implementation and relapse prevention in a group intervention, yet with clear individual differentiation where needed. Because all training sessions were administered by allied health professionals, the Energetic program is implementable in different clinical settings and countries, for different types of NMD. An advantage of Energetic over CBT is that ECM and AET can be administered by regular occupational and physical therapists, whereas CBT must be given by highly trained psychologists. As self-management is an ongoing process in people's lives, short booster sessions or self-help group meetings may potentially prolong the benefits of Energetic as observed in the present study.[36]

The heterogeneity of our study sample may be regarded as a limitation from a mechanistic point of view. There is, however, an increasing body of evidence that the mechanisms underlying chronic fatigue, activity limitations, and participation restrictions share commonalities among various types of chronic medical conditions, independent of their genetic cause or molecular pathophysiology.[10,37,38] Indeed, Menting and colleagues[10] showed that only 11% of the variance in fatigue severity was explained by the type of chronic disease, whereas 55% explained variance was reached when transdiagnostic factors were added to the model, such as sex, age, motivational and concentration problems, pain, sleep disturbances, physical functioning, reduced activity, and lower self-efficacy concerning fatigue. Therefore, in the current study, we deliberately made a next step by moving away from a disease-oriented approach to a patient- and problem-oriented approach. In this perspective, we aimed for functional homogeneity regarding the effect of fatigue in daily life, rather than homogeneity based on disease diagnosis. As homogeneous groups with a single NMD diagnosis are almost impossible to recruit in daily clinical practice, the current study has strong ecological validity and its results are probably better generalizable than the results of NMD-specific studies. Moreover, the applied multifaceted intervention is well feasible in many different settings for patients with various types of NMD and chronic fatigue.

As usual care in this study consisted of a variety of possibly effective interventions, this type of control condition most certainly reduced the contrast between our study groups, rendering the group comparison less specific. Yet, from an ecological point of view, comparing Energetic with usual care fits best with regular clinical practice in most western countries, ensuring a realistic estimation of its effect size.

A limitation of a client-centered criterion-referenced outcome, such as the COPM, is its inherently subjective nature as well as the fact that personal goals may change over time. This latter aspect may have further reduced the observed effect size, since a priority indicated by a patient at baseline may not always be his or her priority at follow-up.[18] Therefore, further research is needed to develop a patient-centered outcome measure that accounts for changes in prioritized activities over time. Future research should also gain insight in the personal factors that contribute to the effects of Energetic in order to optimize patient selection, timing of the intervention, and relapse prevention.

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Nab (occupational therapists), and Tamara Popping (physical therapists) from Radboud University Medical Center. They also thank Anja Horemans and Anke Groenen from the patient support association "Spierziekten Nederland" and Yvonne Cornelissen for help with patient recruitment; Hans Groenewoud for conducting the randomization; Jana Zajec and Bart Kral from Radboud University Medical Center for assistance in the assessments; and Laurien Honing for administrative assistance.

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This was an investigator-initiated study. The sponsors of the study had no role in the study design, patient recruitment, data collection, data analysis, data interpretation, writing of the report, or decision in submitting papers for publication. The corresponding author had full access to all the study data and had the final responsibility for the decision to submit for publication. Funding: Netherlands Organization for Health Research and Development (ZonMW), National Rehabilitation Fund, Centre of Expertise "Sneller Herstel" (HAN), and Dutch FSHD foundation (FSHD Stichting).

## Disclosure

Y. Veenhuizen was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, Centre of Expertise "Sneller Herstel" (HAN), and the Dutch FSHD Foundation. E. Cup was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, Centre of Expertise "Sneller Herstel" (HAN), and the Dutch FSHD Foundation, and reports grants from the Prinses Beatrix Spierfonds. M. Jonker and N. Voet report no disclosures relevant to the manuscript. B. van Keulen was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, and Centre of Expertise "Sneller Herstel" (HAN). D. Maas and A. Heeren report no disclosures relevant to the manuscript. J. Groothuis reports grants from Prinses Beatrix Spierfonds and Fulcrum Therapeutics. B. van Engelen was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, and Centre of Expertise "Sneller Herstel" (HAN), and received grants from the EU Horizon 2020 research and innovation program (Murab), the Netherlands Organization for Scientific Research, the Netherlands Organization for Health Research and Development, Global FSH, Prinses Beatrix Spierfonds, Stichting Spieren voor Spieren, Association Francaise contre les Myopathies, Fulcrum, and the Dutch FSHD Foundation. A. Geurts was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, and Centre of Expertise "Sneller Herstel" (HAN), and received grants from the Netherlands Organization for Scientific Research (ZonMw), the Netherlands Organization for Health Research and Development, Prinses Beatrix Spierfonds, National Rehabilitation Fund, Ipsen, Merz, and Otto Bock. Go to Neurology.org/N for full disclosures.

## Publication history

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## Appendix Authors

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|---|---|---|---|
| **Yvonne Veenhuizen, MSc** | Department of Rehabilitation, Donders Institute for Brain, Cognition and Behaviour, Radboud University Medical Center, Nijmegen, the Netherlands | Author | Conceptualized the study and coordinated funding; undertook and monitored the study with supervision from E.H.C.C., J.T.G., and A.C.H.G., B.G.M.E.; responsible for supervising the therapists and research assistants and recruited patients; did data cleaning and preparations for analysis and interpreted data; wrote the first draft of the manuscript and was responsible for revisions |
| **Edith H.C. Cup, PhD** | Department of Rehabilitation, Donders Institute for Brain, Cognition and Behaviour, Radboud University Medical Center, Nijmegen, the Netherlands | Author | Conceptualized the study and coordinated funding, supervised Y.V. and interpreted data, wrote the first draft of the manuscript and was responsible for revisions |
| **Marianne A. Jonker, PhD** | Department of Health Evidence, Radboud Institute for Health Sciences, Radboud University Medical Center, Nijmegen, the Netherlands | Author | Led the data analysis and wrote the statistical sections, discussed and commented on draft versions |
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## Appendix *(continued)*

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|---|---|---|---|
| **Anita Heeren, MD** | Rehabilitation Center Klimmendaal, Arnhem, the Netherlands | Author | Responsible for the coordination and recruitment in the rehabilitation setting, discussed and commented on draft versions |
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## References


1. Lou JS, Weiss MD, Carter GT. Assessment and management of fatigue in neuromuscular disease. Am J Hosp Palliat Care 2010;27:145–157.
2. Kalkman JS, Schillings ML, van der Werf SP, et al. Experienced fatigue in facioscapulohumeral dystrophy, myotonic dystrophy, and HMSN-I. J Neurol Neurosurg Psychiatry 2005;76:1406–1409.
3. Hibbard JH, Stockard J, Mahoney ER, Tusler M. Development of the Patient Activation Measure (PAM): conceptualizing and measuring activation in patients and consumers. Health Serv Res 2004;39:1005–1026.
4. Voet N, Bleijenberg G, Hendriks J, et al. Both aerobic exercise and cognitive-behavioral therapy reduce chronic fatigue in FSHD: an RCT. Neurology 2014;83:1914–1922.
5. Voet NB, van der Kooi EL, Riphagen II, Lindeman E, van Engelen BG, Geurts AC. Strength training and aerobic exercise training for muscle disease. Cochrane Database Syst Rev 2013:CD003907.
6. Cup EH, Pieterse AJ, Ten Broek-Pastoor JM, et al. Exercise therapy and other types of physical therapy for patients with neuromuscular diseases: a systematic review. Arch Phys Med Rehabil 2007;88:1452–1464.
7. Okkersen K, Jimenez-Moreno C, Wenninger S, et al. Cognitive behavioural therapy with optional graded exercise therapy in patients with severe fatigue with myotonic dystrophy type 1: a multicentre, single-blind, randomised trial. Lancet Neurol 2018;17:671–680.
8. Mathiowetz VG, Matuska KM, Finlayson ML, Luo P, Chen HY. One-year follow-up to a randomized controlled trial of an energy conservation course for persons with multiple sclerosis. Int J Rehabil Res 2007;30:305–313.
9. Blikman LJ, van Meeteren J, Twisk JW, et al. Effectiveness of energy conservation management on fatigue and participation in multiple sclerosis: a randomized controlled trial. Mult Scler 2017;23:1527–1541.
10. Menting J, Tack CJ, Bleijenberg G, et al. Is fatigue a disease-specific or generic symptom in chronic medical conditions? Health Psychol 2018;37:530–543.
11. Veenhuizen Y, Cup EH, Groothuis JT, et al. Effectiveness and cost-effectiveness of a self-management group program to improve social participation in patients with neuromuscular disease and chronic fatigue: protocol of the Energetic study. BMC Neurol 2015;15:58.
12. Geoffrey Moore JLD. Patricia Painter ACSM's Exercise Management for Persons with Chronic Diseases and Disabilities. 4 ed. Champaign: Human Kinetics; 2016.
13. Sauriol A, Brink N, Packer TL. Managing Fatigue: A Six-Week Course for Energy Conservation. Tucson: Therapy Skill Builders; 1995.
14. Cup EH, Pieterse AJ, Knuijt S, et al. Referral of patients with neuromuscular disease to occupational therapy, physical therapy and speech therapy: usual practice versus multidisciplinary advice. Disabil Rehabil 2007;29:717–726.
15. McColl MA, Law M, Baptiste S, Pollock N, Carswell A, Polatajko HJ. Targeted applications of the Canadian Occupational Performance Measure. Can J Occup Ther 2005;72:298–300.
16. Dedding C, Cardol M, Eyssen IC, Dekker J, Beelen A. Validity of the Canadian Occupational Performance Measure: a client-centred outcome measurement. Clin Rehabil 2004;18:660–667.
17. Eyssen IC, Beelen A, Dedding C, Cardol M, Dekker J. The reproducibility of the Canadian Occupational Performance Measure. Clin Rehabil 2005;19:888–894.
18. Sturkenboom IH, Graff MJ, Hendriks JC, et al. Efficacy of occupational therapy for patients with Parkinson's disease: a randomised controlled trial. Lancet Neurol 2014;13:557–566.
19. Kirton A, Ciechanski P, Zewdie E, et al. Transcranial direct current stimulation for children with perinatal stroke and hemiparesis. Neurology 2017;88:259–267.
20. Geerdink Y, Aarts P, van der Burg J, Steenbergen B, Geurts A. Intensive upper limb intervention with self-management training is feasible and promising for older children and adolescents with unilateral cerebral palsy. Res Dev Disabil 2015;43-44:97–105.
21. Vercoulen JH, Swanink CM, Fennis JF, Galama JM, van der Meer JW, Bleijenberg G. Dimensional assessment of chronic fatigue syndrome. J Psychosom Res 1994;38:383–392.
22. Enright PL. The Six-Minute Walk Test. Respir Care 2003;48:783–785.
23. Knak KL, Andersen LK, Witting N, Vissing J. Reliability of the 2- and 6-minute walk tests in neuromuscular diseases. J Rehabil Med 2017;49:362–366.
24. Jong AM, van Nes FA, Lindeboom R. The Dutch Activity Card Sort institutional version was reproducible, but biased against women. Disabil Rehabil 2012;34:1550–1555.
25. Foster ER, Golden L, Duncan RP, Earhart GM. Community-based Argentine tango dance program is associated with increased activity participation among individuals with Parkinson's disease. Arch Phys Med Rehabil 2013;94:240–249.
26. Spinhoven P, Ormel J, Sloekers PP, Kempen GI, Speckens AE, Van Hemert AM. A validation study of the Hospital Anxiety and Depression Scale (HADS) in different groups of Dutch subjects. Psychol Med 1997;27:363–370.
27. Romppel M, Herrmann-Lingen C, Wachter R, et al. A short form of the General Self-Efficacy Scale (GSE-6): development, psychometric properties and validity in an intercultural non-clinical sample and a sample of patients at risk for heart failure. Psychosoc Med 2013;10:Doc01.
28. Van Durme T, Macq J, Jeanmart C, Gobert M. Tools for measuring the impact of informal caregiving of the elderly: a literature review. Int J Nurs Stud 2012;49:490–504.
29. Cup E, Nab N, Maas D, et al. ENERGETIC, a multidisciplinary self-management group program for reconditioning and managing fatigue. Neuromuscul Disord 2012;22:896–897.
30. Carswell A, McColl MA, Baptiste S, Law M, Polatajko H, Pollock N. The Canadian Occupational Performance Measure: a research and clinical literature review. Can J Occup Ther 2004;71:210–222.
31. Schreiner AS, Morimoto T, Arai Y, Zarit S. Assessing family caregiver's mental health using a statistically derived cut-off score for the Zarit Burden Interview. Aging Ment Health 2006;10:107–111.
32. Koopman FS, Brehm MA, Beelen A, et al. Cognitive behavioural therapy for reducing fatigue in post-polio syndrome and in facioscapulohumeral dystrophy: a comparison. J Rehabil Med 2017;49:585–590.
33. Packer TL. Self-management interventions: using an occupational lens to rethink and refocus. Aust Occup Ther J 2013;60:1–2.
34. Liepold A, Mathiowetz V. Reliability and validity of the self-efficacy for performing energy conservation strategies assessment for persons with multiple sclerosis. Occup Ther Int 2005;12:234–249.
35. Moss-Morris R, Norton S. Aerobic exercise, cognitive behavioural therapy and energy conservation management for multiple sclerosis (MS) fatigue: are three trials better than one? Mult Scler 2017;23:1436–1440.
36. Audulv Å. The over time development of chronic illness self-management patterns: a longitudinal qualitative study. BMC Public Health 2013;13:452.
37. Kalkman JS, Schillings ML, Zwarts MJ, van Engelen BG, Bleijenberg G. The development of a model of fatigue in neuromuscular disorders: a longitudinal study. J Psychosom Res 2007;62:571–579.
38. Wokke JH. Fatigue is part of the burden of neuromuscular diseases. J Neurol 2007;254:948–949.

# Self-management program improves participation in patients with neuromuscular disease: A randomized controlled trial

Yvonne Veenhuizen, Edith H.C. Cup, Marianne A. Jonker, et al.

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CORRECTIONS

## Neuroanatomy of pediatric postoperative cerebellar cognitive affective syndrome and mutism

*Neurology*® 2020;94:414. doi:10.1212/WNL.0000000000008668

In the editorial "Neuroanatomy of pediatric postoperative cerebellar cognitive affective syndrome and mutism" by Schmahmann,[1] first published online September 16, 2019, Dr. Albazron's last name was misspelled. It appears correctly in the October 15, 2019, issue. The author and the editorial team regret the error.

### Reference

1. Schmahmann JD. Neuroanatomy of pediatric postoperative cerebellar cognitive affective syndrome and mutism. Neurology 2019;93:693–694.

## A large multicenter study of pediatric myotonic dystrophy type 1 for evidence-based management

*Neurology*® 2020;94:414. doi:10.1212/WNL.0000000000008819

In the article "A large multicenter study of pediatric myotonic dystrophy type 1 for evidence-based management" by Lagrue et al.,[1] the sentence in "Discussion" (p. e861) should have corresponded with the data in figure 1c and read: "In comparison to previous reports, the observed paternal transmission rate was higher than expected (12.7% in the CF, 42% in the IF, and 68.4% in the JF)." The authors regret the error.

### Reference

1. Lagrue E, Dogan C, De Antonio M, et al. A large multicenter study of pediatric myotonic dystrophy type 1 for evidence-based management. Neurology 2019;92:e852–e865.

## Self-management program improves participation in patients with neuromuscular disease

### A randomized controlled trial

*Neurology*® 2020;94:414. doi:10.1212/WNL.0000000000008713

In the article "Self-management program improves participation in patients with neuromuscular disease: A randomized controlled trial" by Veenhuizen et al.,[1] first published online September 30, 2019, the affiliations should have read: From the Departments of Rehabilitation (Y.V., E.H.C.C., N.B.M.V., D.M.M., J.T.G., A.C.H.G.) and Neurology (B.G.M.v.E.), Donders Institute for Brain, Cognition and Behaviour, Radboud University Medical Center, Nijmegen; Department of Health Evidence (M.A.J.), Radboud Institute for Health Sciences, Radboud University Medical Center, Nijmegen; and Rehabilitation Center Klimmendaal (N.B.M.V., B.J.v.K., A.H.), Arnhem, the Netherlands. The affiliations appear correctly in the October 29, 2019, issue. The publisher regrets the error.

### Reference

1. Veenhuizen Y, Cup EHC, Jonker MA, et al. Self-management program improves participation in patients with neuromuscular disease: a randomized controlled trial. Neurology 2019;93:e1720–e1731.

 