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## Evaluation of experiences with a self-management programme for patients with neuromuscular disease and chronic fatigue: A mixed methods evaluation alongside an RCT

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3 **Evaluation of experiences with a self-management programme for patients**  
4 **with neuromuscular disease and chronic fatigue: A mixed methods**  
5 **evaluation alongside an RCT**  
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**ABSTRACT**

**Objective:** To obtain insight into experiences of patients with a neuromuscular disease and chronic fatigue and their healthcare professionals regarding content and delivery of a multidisciplinary out-patient self-management group programme to improve social participation. This will inform future implementation.

**Design:** A mixed method study alongside a randomised controlled trial.

**Setting:** University hospital, rehabilitation centre and community health centre.

**Participants:** 29 patients with a neuromuscular disease and chronic fatigue and 13 healthcare professionals participated in this mixed methods study.

**Intervention:** Multidisciplinary group programme, called Energetic, consisted of a four months intervention with weekly meetings and covered four modules: 1) individually tailored aerobic exercise training; 2) education about aerobic exercise; 3) self-management training in applying energy conservation strategies; and 4) implementation and relapse prevention in daily life.

**Main measures:** Quantitative data was collected by a questionnaire measuring patients' (n=25) satisfaction with the perceived results, content, and delivery of the programme. Qualitative data was collected by individual and focus group interviews to gain insight into the experiences of patients (n=18) and healthcare professionals (n=13) on programme facilitators and barriers.

**Results:** Patients were satisfied with the number and length of the sessions, the different modules, and the therapists. Analysis of the interviews led to five themes: 1) The combination of modules makes a complete picture, 2) The programme is physically and mentally intensive, 3) The group setting is valuable, 4) Small variations in delivery occur in different settings, 5) Therapists are coaches. Suggestions for programme improvement include a combination of face to face and e-health, enhancement of therapists' skills in guiding group interventions, and inclusion of more booster sessions to evaluate and maintain self-management competencies.

**Conclusions:** The Energetic program could be implemented in different health care settings, and group setting, and combination of modules proved to be facilitators for improving self-management.

**Trial registration:** Clinicaltrials.gov NCT02208687.

**Keywords:** energy conservation strategies, evaluation research, exercise aerobic, neuromuscular disease, participation social, rehabilitation.

**ARTICLE SUMMARY**

- A mixed method approach resulted in more insight into experiences of participants and professionals with a self-management group programme using questionnaires and interviews on the content and delivery of the programme.
- To obtain different perspectives, patients that participated in the programme and patients that dropped out were interviewed as well as caregivers and different health care professionals involved in the delivery of the programme.
- Experiences from participants and professionals from three different health care settings were obtained to get insight into barriers and facilitators to implement this programme in these different settings
- Only few partners of participants were interviewed, which may have led to a lack of saturation regarding the partners' perspective
- Different strategies were used to enhance the trustworthiness of the findings; independent researchers were involved in carrying out the interviews, and there was triangulation of data collection methods.

## INTRODUCTION

As there is no cure for most neuromuscular diseases (NMDs), managing the symptoms is essential to participate in daily activities. More than 60% of patients with NMD report fatigue as their most disabling symptom.<sup>1-3</sup> A self-management out-patient group programme, called Energetic, has been developed to improve the social participation and physical endurance of patients with NMD and chronic fatigue.<sup>4</sup> This programme combines aerobic exercise training (AET)<sup>1</sup> education about AET, and energy conservation management (ECM),<sup>5</sup> with relapse prevention and implementation in daily life. It is supervised by trained physical and occupational therapists (figure 1).<sup>4,6</sup> The programme supports patients in acquiring and implementing self-management skills for behavioural change in daily life and for preventing relapse in the long term<sup>4</sup>. The behavioural change techniques include individual goal setting, problem solving, action planning, and feedback from peers.<sup>7-9</sup>

A recent randomised controlled trial (RCT) showed that, compared to usual care, the Energetic programme resulted in a significant improvement of social participation, assessed with the Canadian Occupational Performance Measure (COPM)<sup>1</sup>, and better physical endurance, assessed with the six-minute walking test directly after the intervention and at three- and eleven-months follow-up.<sup>6</sup> The RCT only presented results from quantitative outcome measures, not the patients' and healthcare professionals' experiences with the intervention.

This study, therefore, presents a mixed method approach to the evaluation of the Energetic programme to gain insight in how the delivery and content of the intervention was perceived by patients and healthcare professionals. This evaluation was performed in order to provide suggestions for further improvement and implementation of the Energetic programme in different clinical

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3 settings. The research question was: what are the facilitators and barriers regarding the  
4 content and delivery of the Energetic programme?  
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7  
8 [insert figure 1; Content of the Energetic programme delivered by physical and occupational  
9 therapists]  
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## 16 **METHODS**

### 17 **Study design**

18  
19 We used a mixed methods study design that combined quantitative and qualitative techniques  
20 for the evaluation of experiences of the facilitators and barriers regarding the Energetic  
21 programme.<sup>10-13</sup> A questionnaire was developed to measure satisfaction of patients with the  
22 perceived results, content, and delivery of the programme. Qualitative data were collected to  
23 gain insight into the experiences of patients and healthcare professionals and into facilitators  
24 and barriers regarding the Energetic programme. We used a combination of individual  
25 interviews for in-depth experiences and focus group interviews to stimulate interaction and  
26 discussion among patients.  
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### 44 **Intervention**

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46 Energetic was delivered as a self-management multidisciplinary out-patient rehabilitation  
47 programme in small groups, with a minimum of 4 and a maximum of 8 patients per group. It  
48 was offered by trained physical and occupational therapists at three different clinical settings  
49 in the Netherlands: 1) Radboud University Medical Center (Nijmegen; study coordination), 2)  
50 rehabilitation centre Klimmendaal (Arnhem), and 3) community health centre Buitenlust  
51 (Venray). In all settings, Energetic was delivered during a 4-month period, in which patients  
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3 attended the programme twice a week during the first 9 weeks and once a week during the last  
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5 7 weeks (morning and afternoon programme) (figure 1). In the same time periods, patients  
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7 were requested to perform AET at home once a week (first nine weeks) and twice a week (last  
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9 seven weeks). AET and self-management strategies were tailored to the individual patients as  
10  
11 much as possible. In two sessions, a partner or next of kin was involved. The Energetic  
12  
13 programme consisted of the following modules; 1) AET, 2) education about AET, 3) ECM,<sup>5</sup>  
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15 and 4) relapse prevention and implementation in daily life. Before inclusion the occupational  
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17 therapist assessed the motivation and readiness to change as well as the ability to formulate at  
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19 least three personalized participation goals. Details of the intervention have been reported  
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21 previously.<sup>4 6</sup>  
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### 29 *Training of the therapists*

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31 In each setting one occupational and one physical therapist received a one-day training four  
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33 months before the start of the programme. In addition, a detailed manual with the content and  
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35 schedule for each session was provided to each therapist. The one-day training programme  
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37 focused on: 1) knowledge related to the pathophysiology of different NMDs, 2) the rationale  
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39 for the content of the Energetic programme, 3) the theoretical perspective on self-management  
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41 and behavioural change techniques, and 4) how to organise the Energetic programme in the  
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43 local clinical setting. During the delivery of the programme, all therapists joined three  
44  
45 additional education and discussion meetings. These intervision meetings aimed to facilitate  
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47 the exchange of experiences regarding programme content and delivery among therapists and  
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49 to support the adherence to the programme.  
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### 58 **Participants**

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3 Patients included in the RCT were recruited at the departments of Rehabilitation, Neurology,  
4 and Internal Medicine of the Radboud university medical center, as well as in the other  
5 participating centres. The Dutch patient association for NMD (*'Spierziekten Nederland'*)  
6 facilitated recruitment by providing study information on their website, in their magazine, and  
7 by email. The group who received the Energetic programme consisted of 8 men and 21  
8 women (mean age 52 years, range 20-74 years) with a variety of NMD diagnoses. Detailed  
9 information about the inclusion and exclusion criteria and baseline characteristics has been  
10 reported elsewhere<sup>4 6</sup>. For this evaluation, all patients in the intervention group were asked to  
11 participate in the interviews and to fill in the satisfaction questionnaire. Ten patients and  
12 seven partners or next of kin from the first two groups were asked to participate in an  
13 individual semi-structured interview. Additionally, 19 patients were asked to participate in  
14 focus groups. All healthcare professionals involved in the organisation (secretary),  
15 recruitment (physicians), and delivery of the Energetic programme (occupational therapists  
16 and physical therapists) (n=13) were asked to participate in individual semi-structured  
17 interviews.

## 41 **Data collection**

### 43 *Satisfaction questionnaire*

46 A questionnaire was developed using statements regarding the satisfaction with the results,  
47 the content of the Energetic modules, the frequency and length of the therapeutic sessions, the  
48 organisation, and the therapists that delivered the programme (Appendix A). The  
49 questionnaire contained 42 statements; for 21 statements the level of agreement was measured  
50 with an ordinal four-point rating scale from 'not at all' to 'entirely'; for 18 statements an  
51 ordinal rating scale was used ranging from 1 (not satisfied at all) to 10 (maximally satisfied);  
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3 and three open questions were asked to evaluate the perceived valuable aspects of Energetic.  
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5 Patients were also invited to provide narrative comments on what they would like to improve  
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7 in the programme. All patients received the questionnaire after they had finished the  
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9 programme and were asked to complete it independently and anonymously.  
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### 15 *Individual and focus group interviews*

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18 Semi-structured interview guides with nondirective, open-ended questions were made for the  
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20 interviews<sup>14</sup>. The individual interviews with the patients were held at their homes, four  
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22 months after they had finished the Energetic programme. The healthcare professionals were  
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24 interviewed in their work setting. One therapist was interviewed by videocall. The individual  
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26 interviews with the patients and healthcare professionals were conducted by research  
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28 assistants who were not involved in delivery of the Energetic programme. One research  
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30 assistant led the conversation and the other made notes and observations. The individual  
31  
32 interviews lasted approximately 60 minutes. The focus group interviews with the patients  
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34 were organised at the three different clinical locations. They took place immediately  
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36 following the last session of the programme and lasted 60 to 90 minutes. The focus groups  
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38 were conducted by two research occupational therapists (EN, NN) who were experienced with  
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40 qualitative research and knowledgeable of the Energetic programme, but who were  
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42 uninvolved in the delivery of the programme.  
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49 At the start of the interviews, the aim of the research, the procedures, and the privacy policy  
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51 were explained and there was ample opportunity to ask questions before written informed  
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53 consent was obtained. The patients were interviewed regarding their experiences with the  
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55 content and delivery of the Energetic programme. The healthcare professionals were  
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57 interviewed regarding their experiences with the delivery of the programme (Appendix B).  
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3 Additionally, the logs and notes of the education and discussion meetings with the therapists  
4 were collected for qualitative analysis. Patients (PA), partners or next of kin (NoK) and  
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6 healthcare professionals, including occupational therapists (OT), physical therapists (PT),  
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8 physicians (PHYS), and secretary (SC), were given a number to ensure their anonymity. The  
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10 setting was indicated as Nijmegen (N), Arnhem (A), or Venray (V). For healthcare  
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12 professionals no indication of the setting was provided to ensure anonymity.  
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## 20 **Data analysis**

### 21 *Questionnaires*

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23 We analysed the data from the satisfaction questionnaire using descriptive statistics.<sup>15</sup>  
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27 Statistical analyses were carried out with SPSS version 22.  
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### 33 *Interviews and open questions*

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35 The aim of the analysis was to identify overarching themes regarding facilitators and barriers  
36  
37 related to the content and delivery of the Energetic programme. The analysis process  
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39 consisted of the following steps:<sup>16</sup> 1) individual and focus group interviews were transcribed  
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41 verbatim; 2) qualitative data (transcripts of the individual and focus group interviews, text of  
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43 the open questions of the satisfaction questionnaire, and notes of the therapists' meetings) was  
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45 imported into analysis software for qualitative data (Atlas ti, Version 8.0.34); 3) and was read  
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47 by the first author (YV) to get familiarised; 4) open data coding was conducted by the first  
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49 author (YV) and part of the transcript was also coded by a second author (EC) followed by  
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51 comparison and discussion of the codes (YV/EC) to reach consensus on the coding procedure  
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53 and content; 5) the first (YV) and last author (EC) identified potential categories among the  
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55 initial codes; 6) the potential categories were discussed by members of the research group  
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3 (YV, EC, TS, MNvS) and further grouped into final, main themes related to the research  
4 question; 7) the themes and description of the themes were emailed to all participants and  
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6 were asked if they could identify themselves with these themes. No further comments were  
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8 given on the themes.  
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### 11 12 13 14 15 **Ethical considerations**

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17 All patients gave their written informed consent to participate in the Energetic study.<sup>4</sup>  
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19 Furthermore, all participants in the current study signed an additional informed consent form  
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21 prior to the interviews and questionnaires. Full ethical approval was granted by the regional  
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23 medical ethical committee of Arnhem-Nijmegen (NL47624.091.14) and all participating  
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25 centres granted local approval.  
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## 33 **RESULTS**

### 34 35 **Participants**

#### 36 37 *Patients and partners*

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39 Of the 29 patients in the Energetic programme 25 (86%) completed the satisfaction  
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41 questionnaires. Three patients dropped out of the intervention due to co-morbidity (n=1) or  
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43 experienced too high a burden of the programme (n=2). Ten patients were invited to  
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45 participate in the interviews, of whom four declined due to practical reasons. Thus, six  
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47 patients participated in the individual interviews (of which four patients completed the  
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49 programme and two dropped out). In addition, two partners participated in the individual  
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51 interviews. Of the other 19 patients who were asked to participate in the focus groups, seven  
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53 patients declined to participate for practical reasons. Thus, 12 patients participated in the  
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focus group interviews (see Table 1 for details). Taken together, 18 out of 29 patients took part in the interviews (individual or focus group).

Table 1: Participants in the qualitative interviews.

Participants	Patient (individual interviews) Sex (F/M) Age category diagnoses, work	Patients (focus group) Sex, age category, diagnoses, work	Partners (individual interviews) Sex, age category, work	Healthcare professionals
<b>Rehabilitation center Klimmendaal, Arnhem</b>	1. F, 70-80 years, FSHD, not working 2. F, 60-70 years, IBM, not working	1. F, 40-50 years, FSHD, not working 2. M, 60-70 years, HMSN, working 3. M, 60-70 years, MM, working	1. M, 70-80 years not working	Individual interviews: Rehabilitation physician (n=1) Occupational therapist (n=1) Physical therapist (n=1)
<b>Community health center Buitenlust, Venray</b>	3. F, 60-70 years CPEO, not working, drop out from intervention group	4. F, 60-70 years, MM, not working 5. F, 30-40 years, MM, working 6. M, 60-70 years, HMSN, not working 7. M, 40-50 years, FSHD, Working	-	Individual interviews: Occupational therapist (n=1) Physical therapist (n=1)
<b>Radboud University Medical Center, Nijmegen</b>	4. M, 50-60 years, MM, not working 5. F, 60-70 years, IBM, not working 6. F, 50-60 years, myasthenia gravis, working 7. M, 30-40 years, MM, not working dropped out from intervention group	8. M, 60-70 years, FSHD, working 9. M, 40-50 years, MD, not working 10. F, 30-40 years, MD, working 11. F, 70-80 years, FSHD, working 12. F, 40-50 years, HMSN, not working	2. F, 60-70 years, working	Individual interviews: Rehabilitation physician (n=1) Occupational therapists (n=2) Neurologist (n=1) Internist (n=1) Secretary (n=1) Physical therapist (n=1) Member patient support association (n=1)
<b>Total interviews</b>	N =7	N= 12	N=2	N= 13

FSHD = facioscapulohumeral dystrophy; IBM = inclusion body myositis; MM = mitochondrial myopathy; MD= myotonic dystrophy type 1; CPEO = chronic progressive external ophthalmoplegia; HMSN = hereditary sensory motor neuropathy

### *Healthcare professionals*

All thirteen healthcare professionals involved in the recruitment, organisation and delivery of the Energetic programme were interviewed. One professional was involved in the logistics and planning (secretary), five professionals were involved in the recruitment (four physicians and one representative of the patient association), and seven professionals were involved in the delivery of the programme (three PT and four OT).

### **Satisfaction questionnaire**

The analysis of the satisfaction questionnaire (Table 2) showed that 96% of the patients were entirely or largely satisfied with the results of the intervention. The mean grade of satisfaction with Energetic was 8.7 (SD 1.1) (scale 1-10). Management of the impairments was perceived as “entirely” or “largely” improved by 88% of the patients and the Energetic programme was “largely” (32%) or “entirely” (68%) recommended to others/peers. Regarding the content of Energetic, patients were overall satisfied with the number and length of the sessions, as well as with the therapists and the different modules. In total, 24% of the patients evaluated the total period of 16 weeks as too short, whereas 20% considered the number of sessions in the implementation and relapse prevention module as too high. The sessions on nutrition and work were rated lower than other sessions.

Table 2: Patient satisfaction questionnaire regarding the results, delivery and content of the Energetic programme.

Satisfaction with the results †	Respondents N	Entirely	Largely	Slightly	Not at all
Satisfaction with the intervention results	25	18 (73%)	6 (25%)	1 (2%)	0
Better management of impairments	25	13 (52%)	9 (36%)	3 (12%)	0
Recommendation of Energetic	25	17 (68%)	8 (32%)	0	0
<b>Satisfaction with the number of sessions†</b>		<b>Just right</b>	<b>Too few/too short</b>	<b>Too many/too long</b>	
Per week	25	25 (100%)	0	0	
Aerobic exercise training	25	23 (92%)	1 (4%)	1 (4%)	
Physical education	25	20 (80%)	2 (8%)	3 (12%)	
Energy conservation management	24	21 (84%)	1 (4%)	2 (8%)	
Implementation relapse prevention	25	17 (68%)	3 (12%)	5 (20%)	
Total period (16 weeks)	25	19 (76%)	6 (24%)	0	
<b>Satisfaction with the length of the sessions†</b>					
Fatigue management	24	16 (64%)	6 (24%)	2 (8%)	
Aerobic exercise training	24	18 (72%)	4 (16%)	2 (8%)	
Education about aerobic exercise	24	20 (80%)	2 (8%)	2 (8%)	
<b>Involvement of partner/next of kin in Energetic †</b>	23	21 (84%)	2 (8%)	0	
<b>Rating the content of the sessions ‡</b>		<b>Mean rate 1-10 (SD)</b>			
Energy conservation management	24	7.6 (1.7)			
Aerobic exercise training	24	8.8 (1.1)			
Education on aerobic exercise	24	8.4 (1.0)			
Experience regular sports	25	7.9 (1.4)			
Food and nutrition	25	6.3 (2.4)			
Work/employment	22	6.1(2.5)			
Location/facilities	25	8.4 (1.2)			
Physical therapists: physical training	24	8.9 (1.0)			
Occupational therapist: Energy conservation management	24	8.7 (1.2)			
<b>Total programme Energetic</b>	<b>25</b>	<b>8.7 (1.1)</b>			

† Percentage responding patients with the number and length of sessions. ‡ Mean rating (standard deviation) of the content of the sessions by patients.

## Interviews

The interviews with patients, partners and healthcare professionals resulted in five main themes. Table 3 shows an overview of the main themes, subthemes and quotes. Each theme is described and supported by quotes from the participants.



Table 3: Overview of patients', partners' and healthcare professionals' perspectives regarding the content and delivery of the Energetic programme

Themes and subthemes
<p>1. The combination of modules makes a complete picture.</p> <p>A. The combination of physical training and fatigue management gives insight into one's capacities  <i>"The combination creates a complete picture," (PAN1).</i>  <i>"I could, therefore, still participate in the cake decoration course, if I plan it in as part of my day programme." (PAA5).</i>  <i>"After illness I have to slowly build the physical training programme up to a given point." (PAA5).</i>  <i>"He has learnt to cope better mentally and that was very important for me." (CG2).</i></p> <p>B. Being prepared to change lifestyle is pivotal  <i>"We were all prepared to change things in our lifestyle, to adapt things and to try new things," (PAV2).</i></p> <p>C. Sustainability of implementation in daily life is essential  <i>"A step-by-step guide was provided that could easily be applied practically," (PAN8).</i>  <i>"Good to correct the entrenched deviations and also ask a huge amount of questions," (PAA5).</i></p> <p>D. Sport's participation in one's own environment challenging  <i>"They had already approached clubs before we had finished the Energetic programme," (OT).</i>  <i>"The different sports activities could be placed earlier in the programme, so that I can find out what is good for me." (PAN2).</i></p>
<p>2. The programme is physically and mentally intensive  <i>"The conversation was exhausting, more difficult and more confrontational than expected," (PAA3).</i>  <i>"I had underestimated the time. It cost me a lot more time than expected. This made it difficult to plan in with my home situation." (PAV5).</i>  <i>"All consultations were useful and important for me" (PAA3).</i>  <i>"The Energetic programme is an extremely complex programme to plan and organise" (SC).</i></p>
<p>3. The group setting is valuable  <i>"We learn from each other" (PAV5),</i>  <i>"We encourage each other" (PAA5)</i>  <i>"Within the group, different approaches and viewpoints are heard – it does not always come from the therapist" (OT).</i>  <i>"In future, there needs to be more time for individual questions of all participants, because this time there were two people who dominated time with their questions about looking for work and hobby participation," (OT).</i></p>
<p>4. Small variation in delivery in different settings  <i>"Some of the bicycles are a little heavier, then patients (from other therapists) just had to go on those because my patients need a lighter bike. This was always discussed and ended up not being a problem," (PT).</i>  <i>"I found that planning the sports sessions was always labour intensive and needed a lot of explanation towards management," (PT).</i></p>
<p>5. Therapists are coaches  <i>"allowing us to think outside of the box" (PAA5).</i>  <i>"real interest" (PAV2),</i>  <i>"expertise" (PAA4),</i>  <i>"Guiding a group and encouraging 'change language' is not something you can</i></p>

- learn really quickly and easily in one training session,” (OT)*
- A. Therapists need education
- “It was really stimulating in terms of the learning activities; it was varied in theory and practice in terms of what needed to be done on the programme,” (PT).*
- “You obtain the information, but you can only really engage in conversation about it when you have tried to apply it yourself,” (PT).*
- “Then you can hear from everyone about how it went,” (OT).*

## Themes

### 1. The combination of modules makes a complete picture.

#### *1.A. The combination of physical training and fatigue management gives insight into one's capacities*

An important characteristic of Energetic for both patients and therapists was the combination of the four modules. Improvement of physical fitness, education about AET, applying ECM strategies, and implementation of advice, training and strategies in daily life helped patients to get insight in their energy levels and physical capacities.

*“The combination creates a complete picture.” (PAN1).*

Patients reported that their participation level had increased, because they had become more aware of their possibilities to manage their energy in daily life.

*“I could, therefore, still participate in the cake decoration course, if I plan it in as part of my day programme.” (PAA5).*

The majority of patients reported that their physical fitness had improved. Additionally, most patients had become more aware of their own physical limitations and had gained a better understanding of how they could manage these limitations during physical activities.

*“After illness I have to slowly build the physical training programme up to a given point.” (PAA5).*

The two partners reported that their spouses had learned to better cope with the symptoms.

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3 *“He has learnt to cope better mentally and that was very important for me.” (NoK2).*  
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9 *1.B. Being prepared to change lifestyle is pivotal*

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11 Before the start of the programme both patients and therapists committed themselves to  
12 participation. An intake assessment was held with the patients to determine if they would be  
13 able to participate in the programme for 16 weeks and willing to integrate what they would  
14 learn into their everyday lives. They also identified what they wanted to achieve through  
15 participation in the Energetic programme.  
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23 *“We were all prepared to change things in our lifestyle, to adapt things and to try new*  
24 *things.” (PAV2).*  
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31 *1.C Sustainability of implementation in daily life is essential*

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33 The steady structure in the programme, the translation to practical situations and the  
34 integration in daily routines were experienced as valuable.  
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38 *“A step-by-step guide was provided that could easily be applied practically.” (PAN8).*  
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41 Three months after completion of the programme there was a booster session, which was  
42 valued as positive by most patients.  
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46 *“Good to correct the entrenched deviations [after three months] and also ask a huge*  
47 *amount of questions.” (PAA5).*  
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51 Several patients reported that they would appreciate more booster sessions in the future to be  
52 better able to retain the newly learnt behaviours in everyday life.  
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59 *1.D. Sports participation in one's own environment is challenging*  
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3 An important element of the Energetic programme is the guidance offered to implement  
4 sports activities in everyday life. Therapists reported that most patients actively sought  
5 possibilities for sports participation. Some patients actually joined a sport in their own  
6  
7  
8 environment after completing the programme.  
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13 *“They had already approached clubs before we had finished the Energetic*  
14 *programme.” (OT).*  
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18 However, others reported that they could not find a suitable sports activity. They expressed  
19 the wish for more support in seeking appropriate sports activities in their neighbourhood.  
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22  
23 The sports sessions that were presented as part of the programme were positively valued, but  
24 also difficult to perform. A few patients would have liked the sports sessions to be presented  
25 earlier in the programme, so that they would have had more time to search and implement an  
26  
27  
28 appropriate sport.  
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33 *“The different sports activities could be placed earlier in the programme, so that I can*  
34 *find out what is good for me.” (PAN2).*  
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## 40 2. The programme is physically and mentally intensive

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43 Both patients and healthcare professionals described Energetic as an intensive programme on  
44 many levels. Most patients mentioned the physical training as a factor that contributed to the  
45 intensity. In addition, the mental strain of having to evaluate and reflect on one's own  
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48 behavioural patterns was also experienced as burdensome.  
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53 *“The conversation was exhausting, more difficult and more confrontational than*  
54 *expected.” (PAA3).*  
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3 Some patients reported that the frequency and duration of the sessions were exhausting in the  
4 context of everyday life, whereas others reported that Energetic fitted well within their daily  
5 routine. By some, the travel distance to the programme was mentioned as a stressor.  
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10 *“I had underestimated the time. It costed me a lot more time than expected. This made*  
11 *it difficult to plan in with my home situation.” (PAV5).*  
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15 Nevertheless, most patients reported that they would not like to see any element of the  
16 programme being deleted.  
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20 *“All consultations were useful and important for me.” (PAA3).*  
21  
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23 The healthcare professionals reported the complex planning of the programme within their  
24 work schedule as intensive.  
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28 *“The Energetic programme is an extremely complex programme to plan and*  
29 *organise” (SC).*  
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### 36 3. The group setting is valuable

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38 All patients and healthcare professionals reported that they experienced the group setting as  
39 valuable in order to share experiences, to learn from others, and to motivate each other.  
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43 *“We learn from each other” (PAV5), “We encourage each other” (PAA5) “Within the*  
44 *group, different approaches and viewpoints are heard – it does not always come from*  
45 *the therapist.” (OT).*  
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50 Both therapists and patients reported a group of six patients as optimal to be able to focus on  
51 all personal dilemmas and questions. One patient and one therapist explicitly expressed that  
52 they did not have enough time for individual questions.  
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3 “In future, there needs to be more time for individual questions of all participants,  
4 because this time there were two people who dominated time with their questions  
5 about looking for work and hobby participation.” (OT).  
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#### 10 11 12 13 4. Small variations in delivery occur in different settings

14  
15 The Energetic programme was offered in a variety of clinical settings. Patients and therapists  
16 reported that practical solutions needed to be found at the various locations.  
17

18  
19 “Some of the bicycles are a little heavier. Then patients (from other therapists) just  
20 had to go on those because my patients needed a lighter bike. This was always  
21 discussed and ended up not being a problem.” (PT).  
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28 It also became apparent that the different clinical settings organised certain details differently.  
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30 At the rehabilitations centre, for example, five minutes of Tai Chi was performed by means of  
31 a warm up before the training. The sports sessions were organised at a regular sports complex  
32 outside the centre. The costs hereof were not covered by the medical insurance and therapists  
33 from two settings experienced this as a draw-back.  
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40 “I found that planning the sports sessions was always labour intensive and needed a  
41 lot of explanation towards management.” (PT).  
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#### 48 5. Therapists are coaches

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50 Patients and therapists alike reported that therapists adopted the role of a ‘coach’ during the  
51 programme. The therapists also reported that the collaboration between them (OT and PT)  
52 was important to guide the group well.  
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57 The therapist’s characteristics that patients found important were “expertise” (PAA4), “real  
58 interest” (PAV2), and “allowing us to think outside of the box” (PAA5). A few patients  
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3 reported that they found the guidance from the therapists insufficient and lacking attention  
4 regarding individual differences. Additionally, some therapists reported that supervising an  
5  
6 entire group required a lot of attention.  
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10 *“Guiding a group and encouraging ‘change language’ is not something you can learn*  
11 *really quickly and easily in one training session.” (OT).*  
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#### 14 15 16 17 18 *5.A. Therapists need education* 19

20 The education programme for therapists was experienced as valuable.  
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23 *“It was really stimulating in terms of the learning activities; it was varied in theory*  
24 *and practice in terms of what needed to be done on the programme,” (PT).*  
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28 A few therapists reported that they would have preferred less time between workshop sessions  
29 to prepare for the Energetic programme in their own setting. They liked obtaining the theory  
30 and then being able to implement in daily life what they had learnt.  
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36 *“You obtain the information, but you can only really engage in conversation about it*  
37 *when you have tried to apply it yourself.” (PT).*  
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41 The therapists reported that interaction among peers was important for everyone’s learning  
42 process. The practice experiences were shared during the group supervision.  
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46 *“Then you can hear from everyone about how it went’.” (OT).*  
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## 51 **DISCUSSION**

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54 A mixed methods evaluation of the Energetic programme showed a diverse picture of the  
55 facilitators and barriers related to the content and delivery of this multidisciplinary out-patient  
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57 group intervention for patients with NMD and chronic fatigue.  
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3 The patients' insight in their own capacities and improved participation level was consistent  
4 with the aim of the Energetic programme and with the observed improvement on the primary  
5 outcome of our RCT, the COPM.<sup>6</sup> The COPM measures experienced problems in activities  
6 that are important and meaningful for an individual.<sup>17 18</sup> The choice of the COPM as a primary  
7 outcome fits with the client-centred approach of Energetic and with the impact reported by  
8 patients in this evaluation. Moreover, the perceived improvement of physical fitness reported  
9 by patients was in line with the observed improvement of physical endurance as measured  
10 with the six-minute walking test in our RCT.<sup>6</sup> To measure insight into patient's own  
11 capacities, the general self-efficacy scale (GSES) was used, which showed no group  
12 difference or change over time. However, the GSES is not specifically designed for the self-  
13 efficacy to implement energy conservation strategies. An alternative self-efficacy assessment  
14 developed by Liepold et al.<sup>19</sup> specifically evaluates self-efficacy in performing energy  
15 conservation management strategies and might be a possible valuable measure in future  
16 programme evaluations.

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36 Patients and healthcare professionals reported that the group setting supported the patients to  
37 learn from their own experiences, as well as from each other, with the therapists taking the  
38 role of a coach. Such vicarious experiences, including verbal (social) persuasion, fit well with  
39 Bandura's self-efficacy theory,<sup>20</sup> and are believed to support behavioural change. In addition,  
40 guidance in embedding AET and ECM strategies by self-monitoring behaviour and receiving  
41 feedback from peers may contribute to patients' self-management capacities<sup>9 21</sup>. However, in  
42 this evaluation, some patients reported that, despite this guidance, they found it difficult to  
43 implement exercising at home and maintain the acquired skills in the long term. This  
44 phenomenon has been described by Packer, who emphasised that self-management is an  
45 ongoing process requiring continuous effort and support to gain knowledge, skills and  
46 confidence over time.<sup>22</sup> Additional booster sessions are, therefore, recommended to enable  
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3 trial-and-error practice in a constantly changing context and to receive encouragement from  
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5 peers and knowledgeable healthcare professionals. These booster sessions should focus on the  
6  
7 maintenance of exercising, planning, and pacing in daily life taking into account the  
8  
9 progressive character of the disease and the changing roles and context.  
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12  
13 Patients reported a high willingness to change during the interviews before the start of the  
14  
15 programme, which was probably related to the motivational screening by occupational  
16  
17 therapists before participation. Nevertheless, the Energetic programme was reported to be  
18  
19 physically and mentally intensive and sometimes difficult to schedule within the weekly  
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21 agenda, which also depended on travel distance. This perceived intensity is an important  
22  
23 factor for patients' willingness to participate in Energetic. A way to reduce the intensity is the  
24  
25 use of blended care, for instance combining e-health and face-to-face sessions.<sup>23 24</sup> The recent  
26  
27 developments during the COVID-19 pandemic, for instance the increase in video calls for  
28  
29 regular healthcare, show that e-health can be used in combination with traditional forms of  
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31 care in outpatient rehabilitation.<sup>25</sup>  
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37 The results of this study suggest that Energetic can be delivered in a rehabilitation centre, in a  
38  
39 specialised hospital department, as well as in a primary care setting. Only minor practical  
40  
41 adjustments were necessary per setting. The collaboration among therapists within and  
42  
43 between settings was considered to be a facilitating factor for the delivery of the programme.  
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45 This is in line with the study by van Dongen et al.,<sup>26</sup> who identified facilitating factors for  
46  
47 interdisciplinary collaboration, such as knowing each other well, organisational factors  
48  
49 regarding the delivery of a intervention and professional meetings, and having a shared  
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51 vision. Additionally, they stressed the importance of a team leader who plays a key role in  
52  
53 overseeing the organisation and guiding the team through the developments.<sup>26</sup> Due to the  
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55 complexity in the organisation and planning of the programme we, therefore, suggest that  
56  
57 every clinical setting should assign a team leader to implement Energetic.  
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3 Both therapists and patients experienced that guiding a group requires specific skills for  
4 therapists. Therapist are trained in individual consultations with patients, suggesting that  
5 specific group didactic skills would be helpful to optimize the group interaction within  
6  
7 Energetic Finally, finding finances for the external sports sessions was reported by therapists  
8 as a barrier. Regular sports activities in society are organised outside the healthcare setting  
9 and, thus, are not within the traditional scope of most therapists and not financially  
10 reimbursed by healthcare insurances. For better implementation of this aspect of the Energetic  
11 programme, collaboration with governmental sports organisations and healthcare  
12 professionals working in regular sports domains should be considered.<sup>27</sup>  
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27 We tried to optimise the credibility of our results by including the perspectives of patients,  
28 partners, and healthcare professionals.<sup>28</sup> Furthermore, we used independent interviewers for  
29 the individual and focus group sessions, independent research assistants to establish the  
30 coding structure's validity, and we emailed the themes to all participants and asked if they  
31 could identify themselves with these themes. We have followed different strategies to  
32 enhance the trustworthiness of the findings: triangulation of data collection methods and  
33 triangulation of researchers (use of two researchers for data collection and analysis).  
34  
35 Furthermore, reflective meetings with the research group to discuss the analytical process and  
36 the preliminary and final themes enhanced the credibility of our data.<sup>29 30</sup> Nevertheless, only  
37 two partners of participants participated, which inevitably has led to lack of saturation  
38 regarding the partners' perspective. In addition, the fact that the satisfaction questionnaires  
39 were only filled in by patients who completed the intervention can be considered a  
40 methodological limitation, because it may have led to selection bias. Yet, we gained some  
41 insight also in the experiences of those who dropped out by interviewing two patients that  
42 discontinued the intervention because of its intensity or due to co-morbidities.  
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## CONCLUSION

This mixed methods evaluation was conducted to investigate the experiences of patients and healthcare professionals involved in a multidisciplinary out-patient self-management group programme called Energetic. The aim of this programme is to improve social participation and physical endurance in people with NMD and chronic fatigue. Patients were overall satisfied with the number and length of the sessions, as well as with the therapists and the different modules. Our results indicate that Energetic can be implemented in different clinical settings and that the use of group sessions and using a combination of AET, education about AET, ECM, and daily-life implementation are facilitators for attaining better self-management. Patient suggestions for programme improvement are the use of blended care interventions, inclusion of more booster sessions, and more guidance in seeking appropriate sports activities in the personal environment. As for the therapists, suggested improvements included enhancement of group supervising skills and collaboration between therapists and society or governmental sports organisations.

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## **Ethics approval and consent to participate**

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 centres. All patients provided oral and written informed consent. The trial was registered at [clinicaltrials.gov](http://clinicaltrials.gov) (NCT02208687).

## **Availability of data and material**

All data used and analysed during the current study are available from the corresponding author on reasonable request until 2026.

## **Declaration of Conflicting Interests**

YV was sponsored by the Netherlands Organization for Health Research and Development (ZonMw), National Rehabilitation Fund, and Centre of Expertise ‘Sneller Herstel’ (HAN) and the Dutch FSHD Foundation.

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2  
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4  
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6  
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34  
35 TS, MJLG, and MNvS declare that there is no conflict of interest.  
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#### 40 **Patients and public partnership**

41  
42 The Energetic programme is developed for and with patients in the outpatient’s rehabilitation  
43  
44 clinic of the department rehabilitation Radboudumc. Patients were not involved in  
45  
46 development of this mixed method evaluation on the Energetic programme. However, the  
47  
48 experiences of patients and health professionals were carefully selected in this study to  
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50 improve the intervention and further implementation.  
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#### 56 **Authors’ contributions**

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2  
3 YV, EHCC, BGMvE and ACHG conceptualised the study and coordinated funding. YV  
4  
5 undertook and monitored the study with supervision from EHCC, JTG, ACHG, TS and  
6  
7 MNvS. YV was responsible for supervising the therapists, research assistants, and recruited  
8  
9 patients. YV and EHCC led the data analysis. YV, EHCC, TS and MNvS interpreted data.  
10  
11  
12 YV and EHCC, TS and MNvS wrote the first draft of the manuscript and were responsible for  
13  
14 revisions. YV, EHCC, TS, MJLG, MNvS, BGMvE and ACHG discussed and commented on  
15  
16 draft versions. All authors approved the final version.  
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## Tables/figures

Table 2: Participants in the qualitative interviews.

FSHD = facioscapulohumeral dystrophy; IBM = inclusion body myositis; MM mitochondrial myopathy; MD= myotonic dystrophy type 1

Table 2: Patient satisfaction questionnaire regarding the perceived delivery and content of the Energetic programme.

† Percentage responding patients with the number and length of sessions. ‡ Mean rating (standard deviation) of the content of the sessions by patients.

Table 3: Overview of patients', partners' and healthcare professionals' perspectives regarding the content and delivery of the Energetic programme

Figure 1: Content of the Energetic programme delivered by physical therapists and occupational therapists.

**A: Satisfaction questionnaire****Planning and organization**

<b>Question</b>	<b>Answer options</b>
Was the total number of days per week adequate for you?	Just right Too few/too short Too many/too long
Was the total number of physical training sessions adequate for you?	Just right Too few/too short Too many/too long
Was the total number of physical education meetings adequate for you?	Just right Too few/too short Too many/too long
Was the total number of sessions for energy conservation management adequate for you?	Just right Too few/too short Too many/too long
Was the total number of sessions for implementation and relapse prevention adequate for you?	Just right Too few/too short Too many/too long
What did you think of the length of the period (16 weeks) in which the treatments took place?	Just right Too few/too short Too many/too long
How do you rate the location and facilities?	Rate 1= extremely bad; 10= extremely good

**Module Energy conservation management**

<b>Question</b>	<b>Answer options</b>
How do you rate session 1: Importance of rest?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 2: Communication, postures and positioning?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 3: Practical situations?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 4: Priorities/standards/norms and values and analysis/adaptation of activities?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 5: Balance in your schedule?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 6: Evaluation and future plans?	Rate 1= extremely useless; 10= extremely useful
How do you rate the length of session's energy conservation management?	Just right Too few/too short Too many/too long
How do you rate the way the sessions were supervised by the occupational therapist?	Rate 1= extremely useless; 10= extremely useful

**Module Aerobic exercise training**

<b>Question</b>	<b>Answer options</b>
How do you rate the added value/use of the physical training?	Rate 1= extremely useless; 10= extremely useful
How do you rate the way in which the sessions were supervised by the physical therapist?	Rate 1= extremely useless; 10= extremely useful
What did you think of the length of the physical training sessions?	Just right Too few/too short Too many/too long

**Module physical education**

<b>Question</b>	<b>Answer options</b>
How do you rate session 1: Introduction and training theory	Rate 1= extremely useless; 10= extremely useful

How do you rate session 2: Effects of training	Rate 1= extremely useless; 10= extremely useful
How do you rate session 3: Completion of the training & preparation of a training schedule	Rate 1= extremely useless; 10= extremely useful
What did you think of the length of the physical education sessions?	Just right Too few/too short Too many/too long
How do you rate the way in which the sessions were supervised by the physical therapist?	Rate 1= extremely useless; 10= extremely useful

### Module implementation and relapse prevention

Question	Answer options
What did you think of the extent to which your next of kin or partner was involved in the programme?	Just right Too few/too short Too many/too long
How do you rate the various sports sessions?	Rate 1= extremely useless; 10= extremely useful
How do you rate the dietetics/nutrition session?	Rate 1= extremely useless; 10= extremely useful
How do you rate employment session?	Rate 1= extremely useless; 10= extremely useful

### Therapists

Question	Answer options
Did you find the occupational therapist competent?	No, not at all A little Largely so Yes, entirely
Did the occupational therapist give advice that is appropriate and useful for your situation?	No, not at all A little Largely so Yes, entirely
Did you find the physical therapist competent?	No, not at all A little Largely so Yes, entirely
Did the physical therapist give advice that is suitable and useful for your situation?	No, not at all A little Largely so Yes, entirely

### Treatment

Question	Answer options
Did the occupational therapist treat you politely and with respect?	No, not at all A little Largely so Yes, entirely
Did the occupational therapist make you feel at ease?	No, not at all A little Largely so Yes, entirely
Did the physical therapist treat you politely and with respect?	No, not at all A little Largely so Yes, entirely
Did the physical therapist ensure that you felt at ease?	No, not at all A little Largely so Yes, entirely

## Results

Question	Answer options
Are you satisfied with the results of the Energetic programme?	No, not at all A little Largely so Yes, entirely
Can the Energetic programme help you deal with your limitations and/or problems better than before?	No, not at all A little Largely so Yes, entirely

### Overall satisfaction

Question	Answer options
Suppose you have a good friend who is in the same situation as you. Would you recommend this friend to participate in the Energetic programme?	Yes, absolutely Yes, maybe No I don't know if I would do that
How do you rate your satisfaction with the Energetic programme?	Rate 1= extremely bad; 10= extremely good

### Open questions:

- The most valuable for me was:
- If I could change the programme, I would change....
- Space for comments on the Energetic programme

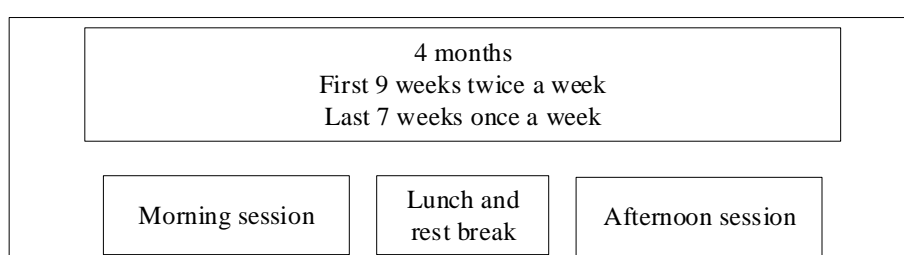
### Appendix B: Topic lists interviews.

Individual interviews with patients and partners	
Perceived impact	What is your experience with Energetic?
Content of Energetic	What were the positive and negative aspects of the content of Energetic? Elements: Experience with Energetic, different modules, frequency, lengths, construction, group, goal setting, next of kin's involvement, influence on home situation, trainers
Organisation	What were the facilitators and barriers in the delivery and organisation of Energetic? Elements: location, accessibility, travel time, facilities,
Focus group with patients	
	How did you experience the quality of: <ul style="list-style-type: none"> <li>• Content of the sessions</li> <li>• Working methods</li> <li>• Education</li> <li>• Individual goals</li> <li>• Group setting</li> <li>• Trainers</li> <li>• Organization</li> <li>• involvement of next of kin/partners</li> </ul> <p>Creative questions regarding the benefit of Energetic. For instance:</p> <ul style="list-style-type: none"> <li>• If you could make a commercial about Energetic for the health insurance, what would you mention in this commercial?</li> <li>• Suppose you are in the waiting room in the hospital and would meet a peer,</li> </ul>

	<p>what would you tell him/her about Energetic?</p> <ul style="list-style-type: none"> <li>• Suppose in one-month time you are back with the rehabilitation physician who referred you to Energetic, what will you tell him/her about the programme?</li> <li>• What components should stay in Energetic?</li> </ul> <p>Barriers:</p> <ul style="list-style-type: none"> <li>• If the health insurance company forces you to adapt the programme in order to reduce costs, what components could be deleted from Energetic?</li> <li>• What components could be altered or deleted from Energetic?</li> </ul> <p>Addition:</p> <ul style="list-style-type: none"> <li>• For instance: if money is no issue for Energetic, what would you add to the programme?</li> <li>• Is there some component that you missed in Energetic?</li> <li>• If you must describe your experience with Energetic in a few words, what would that be?</li> </ul>
Individual interviews with healthcare professionals	
All healthcare professionals	General information: name, function in relation to Energetic.
Healthcare professionals involved in the delivery	<p>How was the starting phase (implementation) of the programme?</p> <p>What were the facilitators and barriers for Energetic at your location?</p> <p>What is needed in the programme to implement it nationally?</p> <p>Elements: provided means for implementation, instruction manual, education, meetings, time investment, what should be changed or altered in the starting phase of Energetic (implementation), guidance as a therapist, communication in the programme (in the setting and between the different settings)</p>
Healthcare professionals involved in the recruitment	<p>Can you describe how the recruitment took place?</p> <p>What is needed to recruit patients for Energetic?</p> <p>Can you tell us something about the time investment for recruitment?</p>
Healthcare professionals involved in the organisation	<p>Can you describe how the organisation took place?</p> <p>What means and facilities were necessary to organise Energetic and how did this influence the delivery of the programme?</p>

**Inclusion criteria for the Energetic programme:**

- Patients with a NMD and chronic fatigue influencing their social participation
- Motivation and readiness to change explored by motivational interviewing
- No depressive symptoms or other psychiatric or cognitive symptoms as judged by a psychologist
- Formulation of at least three personalized goals
- Physical capacity to participate in aerobic exercise training based on a cardiopulmonary test.



<p><b>Aerobic exercise training</b></p> <p>9 weeks; 2 times weekly (18 sessions)</p> <p>7 weeks; once a week (7 sessions)</p> <p>Duration: 90 minutes Trainer: Physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Individually tailored exercise training at 50-70% of the maximum heart rate, guided by a cardiac rhythm monitor</li> <li>- Fine tuning takes place based on the recovery rate</li> <li>- Different exercises depending on the reference and motor abilities of the patients.</li> </ul>	<p><b>Energy conservation management[5]</b></p> <p>Weeks 2-8 and 10 (8 sessions)</p> <p>Duration; 90 minutes Trainer: Occupational therapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>-Benefits of rest</li> <li>- Effectively Communicating with the social environment</li> <li>- Applying principles of proper body mechanics and ergonomics</li> <li>- Adequately modifying the personal environment</li> <li>- Analysing and adjusting individual activities</li> <li>- Setting priorities</li> <li>-Finding an activity-rest balance over the entire day and week</li> <li>- Setting short-term and long-term goals and action planning</li> </ul>
<p><b>Education about aerobic exercise</b></p> <p>3 weeks; once a week (3 sessions)</p> <p>Duration: 60 minutes Trainer: Physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Attaining an adequate training stimulus</li> <li>- The need to rest and recuperate</li> <li>- Designing and adhering to a feasible training programme</li> <li>- Prevention of overtraining and relapse</li> <li>- Management physical training after relapse</li> </ul>	<p><b>Relapse prevention and implementation and in daily life</b></p> <p>Weeks 5-13 and 16 (8 sessions)</p> <p>Duration training: 60 minutes Trainer: Occupational therapist and physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Education by a dietician</li> <li>- NMD and fatigue regarding work</li> <li>- Two sessions with the partners/next of kin</li> <li>- Different types of exercise to explore (swimming, Nordic walking, yoga, body work-out, dancing)</li> <li>- Explore possibilities for exercising in patients own environment</li> </ul>

**The TIDieR (Template for Intervention Description and Replication) Checklist\*:**

Information to include when describing an intervention and the location of the information

Item number	Item	Where located **	
		Primary paper (Page or appendix number)	Other † (details)
1.	<b>BRIEF NAME</b> Provide the name or a phrase that describes the intervention.	Page 2, 5, 6	_____
2.	<b>WHY</b> Describe any rationale, theory, or goal of the elements essential to the intervention.	Page 4	_____
3.	<b>WHAT</b> Materials: Describe any physical or informational materials used in the intervention, including those provided to participants or used in intervention delivery or in training of intervention providers. Provide information on where the materials can be accessed (e.g. online appendix, URL).	Page 5-7	_____
4.	Procedures: Describe each of the procedures, activities, and/or processes used in the intervention, including any enabling or support activities.	Figure 1, page 5-	_____
5.	<b>WHO PROVIDED</b> For each category of intervention provider (e.g. psychologist, nursing assistant), describe their expertise, background and any specific training given.	Figure 1, page 5-	_____
6.	<b>HOW</b> Describe the modes of delivery (e.g. face-to-face or by some other mechanism, such as internet or telephone) of the intervention and whether it was provided individually or in a group.	Figure 1, page 5-	Veenhuizen Y, Cup EH, Groothuis JT, et al. Effectiveness and cost-effectiveness of a self-management group program to



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<p><b>WHERE</b></p> <p>7. Describe the type(s) of location(s) where the intervention occurred, including any necessary infrastructure or relevant features.</p>	<p>improve social participation in patients with neuromuscular disease and chronic fatigue: protocol of the Energetic study. <i>BMC Neurol</i> 2015; 15: 58. DOI: 10.1186/s12883-015-0314-4.</p> <p>page 5,6</p> <p>Veenhuizen 2015</p>
<p><b>WHEN and HOW MUCH</b></p> <p>8. Describe the number of times the intervention was delivered and over what period of time including the number of sessions, their schedule, and their duration, intensity or dose.</p> <p><b>TAILORING</b></p> <p>9. If the intervention was planned to be personalised, titrated or adapted, then describe what, why, when, and how.</p> <p><b>MODIFICATIONS</b></p> <p>10.* If the intervention was modified during the course of the study, describe the changes (what, why, when, and how).</p> <p><b>HOW WELL</b></p> <p>11. Planned: If intervention adherence or fidelity was assessed, describe how and by whom, and if any strategies were used to maintain or improve fidelity, describe them.</p> <p>12.* Actual: If intervention adherence or fidelity was assessed, describe the extent to which the intervention was delivered as planned.</p>	<p>figure 1,</p> <p>page 20-23</p> <p>page 20-23</p> <p>page 20-23</p> <p>Veenhuizen 2015</p> <p>Veenhuizen 2015</p>

1 \*\* **Authors** - use N/A if an item is not applicable for the intervention being described. **Reviewers** – use ‘?’ if information about the element is not reported/not  
2 sufficiently reported.  
3

4 † If the information is not provided in the primary paper, give details of where this information is available. This may include locations such as a published protocol  
5 or other published papers (provide citation details) or a website (provide the URL).  
6

7 ‡ If completing the TIDieR checklist for a protocol, these items are not relevant to the protocol and cannot be described until the study is complete.

8 \* We strongly recommend using this checklist in conjunction with the TIDieR guide (see *BMJ* 2014;348:g1687) which contains an explanation and elaboration for each item.  
9

10 \* The focus of TIDieR is on reporting details of the intervention elements (and where relevant, comparison elements) of a study. Other elements and methodological features of  
11 studies are covered by other reporting statements and checklists and have not been duplicated as part of the TIDieR checklist. When a **randomised trial** is being reported, the  
12 TIDieR checklist should be used in conjunction with the CONSORT statement (see [www.consort-statement.org](http://www.consort-statement.org)) as an extension of **Item 5 of the CONSORT 2010 Statement**.  
13 When a **clinical trial protocol** is being reported, the TIDieR checklist should be used in conjunction with the SPIRIT statement as an extension of **Item 11 of the SPIRIT 2013**  
14 **Statement** (see [www.spirit-statement.org](http://www.spirit-statement.org)). For alternate study designs, TIDieR can be used in conjunction with the appropriate checklist for that study design (see  
15 [www.equator-network.org](http://www.equator-network.org)).  
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# BMJ Open

## A mixed methods evaluation of a self-management group programme for patients with neuromuscular disease and chronic fatigue

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<b>Primary Subject Heading</b>:	Rehabilitation medicine
Secondary Subject Heading:	Neurology
Keywords:	Neuromuscular disease < NEUROLOGY, REHABILITATION MEDICINE, SPORTS MEDICINE

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1 **A mixed methods evaluation of a self-management group programme for**  
2 **patients with neuromuscular disease and chronic fatigue**

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## 1 ABSTRACT

2 **Objective:** To obtain insight into experiences of patients with a neuromuscular disease  
3 and chronic fatigue and their healthcare professionals regarding content and delivery of  
4 a multidisciplinary out-patient self-management group programme to improve social  
5 participation. This will inform future implementation.

6 **Design:** A mixed method study alongside a randomised controlled trial.

7 **Setting:** University hospital, rehabilitation centre and community health centre.

8 **Participants:** 29 patients with a neuromuscular disease and chronic fatigue and 13 healthcare  
9 professionals participated in this mixed methods study.

10 **Intervention:** Multidisciplinary group programme, called Energetic, consisted of a four  
11 months intervention with weekly meetings and covered four modules: 1) individually tailored  
12 aerobic exercise training; 2) education about aerobic exercise; 3) self-management training in  
13 applying energy conservation strategies; and 4) implementation and relapse prevention in  
14 daily life.

15 **Main measures:** Quantitative data was collected by a questionnaire measuring patients'  
16 (n=25, all completed the programme) satisfaction with the perceived results, content, and  
17 delivery of the programme. Qualitative data was collected by individual and focus group  
18 interviews to gain insight in the experiences of patients (n=18), next of kin (n=2), and  
19 healthcare professionals (n=13) with facilitators and barriers to programme implementation.

20 **Results:** Patients were satisfied with the number and length of the sessions, the different  
21 modules, and the therapists. Analysis of the interviews led to five themes: 1) The combination  
22 of modules makes a complete picture, 2) The programme is physically and mentally intensive,  
23 3) The group setting is valuable, 4) Small variations in delivery occur in different settings, 5)  
24 Therapists are coaches. Suggestions for programme improvement include a combination of  
25 face to face and e-health, enhancement of therapists' skills in guiding group interventions, and  
26 inclusion of more booster sessions to evaluate and maintain self-management competencies.

27 **Conclusions:** The Energetic program could be implemented in different healthcare settings  
28 and group settings, and a combination of modules proved to be a facilitator for improving  
29 self-management.

30 **Trial registration:** Clinicaltrials.gov NCT02208687.

31 **Keywords:** energy conservation strategies, evaluation research, exercise aerobic,  
32 neuromuscular disease, participation social, rehabilitation.

## ARTICLE SUMMARY

### Strengths and limitations of this study

- A mixed method approach resulted in more insight in experiences of participants and professionals with a self-management group programme using questionnaires and interviews on the content and delivery of the programme.
- To obtain different perspectives, patients that participated in the programme and those that dropped out were interviewed as well as caregivers and different healthcare professionals involved in the delivery of the programme.
- Experiences from participants and professionals from three different healthcare settings were obtained to get insight in barriers and facilitators to implement the programme in these different settings.
- Only few partners of participants were interviewed, which may have led to a lack of saturation regarding the partners' perspective.
- Different strategies were used to enhance the trustworthiness of the findings; independent researchers were involved in carrying out the interviews, and there was triangulation of data collection methods.

## 1 INTRODUCTION

2 As there is no cure for most neuromuscular diseases (NMDs), managing the symptoms is  
3 essential for patients with NMD to participate in daily activities. More than 60% of patients  
4 with NMD report fatigue as their most disabling symptom.<sup>1-3</sup> A self-management out-patient  
5 group programme, called Energetic, has been developed to improve the social participation  
6 and physical endurance of patients with NMD and chronic fatigue.<sup>4</sup> This programme  
7 combines aerobic exercise training (AET)<sup>1</sup> education about AET, and energy conservation  
8 management (ECM),<sup>5</sup> with relapse prevention and implementation in daily life. It is  
9 supervised by trained physical and occupational therapists (figure 1).<sup>4,6</sup> The programme  
10 supports patients in acquiring and implementing self-management skills for behavioural  
11 change in daily life and for preventing relapse in the long term<sup>4</sup>. The behavioural change  
12 techniques include individual goal setting, problem solving, action planning, and feedback  
13 from peers.<sup>7-9</sup>

14 A recent randomised controlled trial (RCT) showed that, compared to usual care, the  
15 Energetic programme resulted in a significant improvement of social participation, assessed  
16 with the Canadian Occupational Performance Measure (COPM)<sup>1</sup>, and better physical  
17 endurance, assessed with the six-minute walking test directly after the intervention and at  
18 three- and eleven-months follow-up.<sup>6</sup> The RCT only presented results from quantitative  
19 outcome measures, not the patients' and healthcare professionals' experiences with the  
20 intervention.

21 This study, therefore, presents a mixed method approach to the evaluation of the Energetic  
22 programme to gain insight in the perceived satisfaction of patients and healthcare  
23 professionals with the programme and the factors influencing the intervention. This  
24 evaluation was performed in order to provide suggestions for improvement of the content and  
25 the delivery of the intervention, the perceived impact, patient selection, timing of the



1 intervention, and for further improvement and implementation of the Energetic programme in  
2 different clinical settings. The research questions were: what is the perceived satisfaction with  
3 the Energetic programme and what are the facilitators and barriers regarding the impact,  
4 content and delivery of the Energetic programme?

5 [insert figure 1; Content of the Energetic programme delivered by physical and occupational  
6 therapists]

## 8 **METHODS**

### 9 **Study design**

10 We used a mixed methods study design that combined quantitative and qualitative techniques  
11 for the evaluation of experiences of the facilitators and barriers regarding the Energetic  
12 programme.<sup>10-13</sup> A questionnaire was developed to measure satisfaction of patients with the  
13 perceived results, content, and delivery of the programme. Qualitative data were collected to  
14 gain insight into the experiences of patients and healthcare professionals and into facilitators  
15 and barriers regarding the Energetic programme. We used a combination of individual  
16 interviews for in-depth experiences and focus group interviews to stimulate interaction and  
17 discussion among patients.

### 19 **Intervention**

20 Energetic was delivered as a self-management multidisciplinary out-patient rehabilitation  
21 programme in small groups, with a minimum of 4 and a maximum of 8 patients per group. It  
22 was offered by trained physical and occupational therapists at three different clinical settings  
23 in the Netherlands: 1) Radboud University Medical Center (Nijmegen; study coordination), 2)

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3 1 rehabilitation centre Klimmendaal (Arnhem), and 3) community health centre Buitenlust  
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5 2 (Venray). In all settings, Energetic was delivered during a 4-month period, in which patients  
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7 3 attended the programme twice a week during the first 9 weeks and once a week during the last  
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9 4 7 weeks (morning and afternoon programme) (figure 1). In the same time periods, patients  
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11 5 were requested to perform AET at home once a week (first nine weeks) and twice a week (last  
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13 6 seven weeks). AET and self-management strategies were tailored to the individual patients as  
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15 7 much as possible. In two sessions, a partner or next of kin was involved. The Energetic  
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17 8 programme consisted of the following modules; 1) AET, 2) education about AET, 3) ECM,<sup>5</sup>  
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19 9 and 4) relapse prevention and implementation in daily life. Before inclusion the occupational  
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21 10 therapist assessed the motivation and readiness to change as well as the ability to formulate at  
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23 11 least three personalized participation goals. Details of the intervention have been reported  
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25 12 previously.<sup>4 6</sup>  
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#### 34 *Training of the therapists*

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36 15 In each setting one occupational and one physical therapist received a one-day training four  
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38 16 months before the start of the programme. In addition, a detailed manual with the content and  
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40 17 schedule for each session was provided to each therapist. The one-day training programme  
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42 18 focused on: 1) knowledge related to the pathophysiology of different NMDs, 2) the rationale  
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44 19 for the content of the Energetic programme, 3) the theoretical perspective on self-management  
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46 20 and behavioural change techniques, and 4) how to organise the Energetic programme in the  
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48 21 local clinical setting. During the delivery of the programme, all therapists joined three  
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50 22 additional education and discussion meetings. These intervision meetings aimed to facilitate  
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52 23 the exchange of experiences regarding programme content and delivery among therapists and  
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54 24 to support the adherence to the programme.  
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## 1 **Participants**

2 Patients included in the RCT were recruited at the departments of Rehabilitation, Neurology,  
3 and Internal Medicine of the Radboud university medical center, as well as in the other  
4 participating centres. The Dutch patient association for NMD (*'Spierziekten Nederland'*)  
5 facilitated recruitment by providing study information on their website, in their magazine, and  
6 by email. The group who received the Energetic programme consisted of 8 men and 21  
7 women (mean age 52 years, range 20-74 years) with a variety of NMD diagnoses  
8 (facioscapulohumeral dystrophy, inclusion body myositis, mitochondrial myopathy). Detailed  
9 information about the inclusion and exclusion criteria and baseline characteristics has been  
10 reported elsewhere<sup>4,6</sup>. For this evaluation, all patients in the intervention group were asked to  
11 fill in the satisfaction questionnaire. Ten consecutive patients and seven partners or next of  
12 kin from the first two Energetic groups were asked by email to participate in an individual  
13 semi-structured interview. Additionally, 19 patients were asked face-to-face during the  
14 intervention to participate in focus groups. All healthcare professionals involved in the  
15 organisation (secretary), recruitment (physicians), and delivery of the Energetic programme  
16 (occupational therapists and physical therapists) (n=13) were asked to participate in individual  
17 semi-structured interviews.

## 19 **Data collection**

### 20 *Satisfaction questionnaire*

21 A questionnaire was developed using statements regarding the satisfaction with the results,  
22 the content of the Energetic modules, the frequency and length of the therapeutic sessions, the  
23 organisation, and the therapists that delivered the programme (Appendix A). The  
24 questionnaire contained 42 statements; for 21 statements the level of agreement was measured

1 with an ordinal four-point rating scale from 'not at all' to 'entirely'; for 18 statements an  
2 ordinal rating scale was used ranging from 1 (not satisfied at all) to 10 (maximally satisfied);  
3 and three open questions were asked to evaluate the perceived valuable aspects of Energetic.  
4 Patients were also invited to provide narrative comments on what they would like to improve  
5 in the programme. All patients received the questionnaire after they had finished the  
6 programme and were asked to complete it independently and anonymously.

### 7 8 *Individual and focus group interviews*

9 Semi-structured interview guides with nondirective, open-ended questions were made for the  
10 interviews<sup>14</sup>. The individual interviews with the patients were held at their homes, four  
11 months after they had finished the Energetic programme. The healthcare professionals were  
12 interviewed in their work setting. One therapist was interviewed by videocall. The individual  
13 interviews with the patients and healthcare professionals were conducted by research  
14 assistants who were not involved in delivery of the Energetic programme. One research  
15 assistant led the conversation and the other made notes and observations. The individual  
16 interviews lasted approximately 60 minutes. The focus group interviews with the patients  
17 were organised at the three different clinical locations. They took place immediately  
18 following the last session of the programme and lasted 60 to 90 minutes. The focus groups  
19 were conducted by two research occupational therapists (EN, NN) who were experienced with  
20 qualitative research and knowledgeable of the Energetic programme, but who were  
21 uninvolved in the delivery of the programme.

22 At the start of the interviews, the aim of the research, the procedures, and the privacy policy  
23 were explained and there was ample opportunity to ask questions before written informed  
24 consent was obtained. The patients were interviewed regarding their experiences with the

1 content and delivery of the Energetic programme. The healthcare professionals were  
2 interviewed regarding their experiences with the delivery of the programme (Appendix B).  
3 Additionally, the logs and notes of the education and discussion meetings with the therapists  
4 were collected for qualitative analysis. Patients (PA), partners or next of kin (NoK) and  
5 healthcare professionals, including occupational therapists (OT), physical therapists (PT),  
6 physicians (PHYS), and secretary (SC), were given a number to ensure their anonymity. The  
7 setting was indicated as Nijmegen (N), Arnhem (A), or Venray (V).

## 9 **Data analysis**

### 10 *Questionnaires*

11 We analysed the data from the satisfaction questionnaire using descriptive statistics.<sup>15</sup>

12 Statistical analyses were carried out with SPSS version 22.

### 14 *Interviews and open questions*

15 The aim of the constant comparative analysis was to identify overarching themes regarding  
16 facilitators and barriers related to the content and delivery of the Energetic programme. The  
17 analysis process consisted of the following steps:<sup>16</sup> 1) individual and focus group interviews  
18 were transcribed verbatim; 2) qualitative data (transcripts of the individual and focus group  
19 interviews, text of the open questions of the satisfaction questionnaire, and notes of the  
20 therapists' meetings) was imported into analysis software for qualitative data (Atlas ti,  
21 Version 8.0.34); 3) and was read by the first author (YV) to get familiarised; 4) open data  
22 coding was conducted by the first author (YV) and part of the transcript was also coded by a  
23 second author (EC) followed by comparison and discussion of the codes (YV/EC) to reach  
24 consensus on the coding procedure and content. In total 706 codes were found in the open

1 coding process; 5) the first (YV) and last author (EC) identified potential 14 categories among  
2 the initial codes; 6) the potential categories were discussed by members of the research group  
3 (YV, EC, TS, MNvS) and further grouped into final, main themes related to the research  
4 question; 7) the themes and description of the themes were emailed to all participants and  
5 they were asked if they could identify themselves with these themes. No further comments  
6 were given on the themes. The COREQ checklist was used for reporting the qualitative data.

## 8 **Patient and public Involvement**

9 Patients of our outpatient rehabilitation clinic (at Radboud University Medical Centre) were  
10 involved in designing the Energetic programme. In a pilot study, qualitative research was  
11 conducted to improve the elements of the Energetic programme<sup>17</sup>. The Energetic programme  
12 primarily addressed the patients' main research priority (i.e. fatigue) as indicated by the  
13 research priorities of patients with NMD in general<sup>18</sup>. No patient advisors were involved in  
14 designing the research questions or in the recruitment of this study. The Dutch NMD patient  
15 support association was involved in the recruitment of the participants in the RCT. This  
16 mixed methods evaluation gives insight in the experiences of patients regarding the Energetic  
17 programme, which also includes the burden of the intervention. The results from the  
18 individual and focus group interviews were sent to the participants and they were asked if  
19 they could identify themselves with the collected themes. The results of this study will be  
20 presented at congresses of the Dutch NMD patient support association and in their journals,  
21 which are sent to all members and are written in understandable language.

## 23 **Ethical considerations**

1 All patients gave their written informed consent to participate in the Energetic study.<sup>4</sup>  
 2 Furthermore, all participants in the current study signed an additional informed consent form  
 3 prior to the interviews and questionnaires. Full ethical approval was granted by the regional  
 4 medical ethical committee of Arnhem-Nijmegen (NL47624.091.14) and all participating  
 5 centres granted local approval.

## 7 RESULTS

### 8 Participants

#### 9 *Patients and partners*

10 Of the 29 patients in the Energetic programme 25 (86%) completed the satisfaction  
 11 questionnaires. Three patients dropped out of the intervention due to co-morbidity (n=1) or  
 12 experienced too high a burden of the programme (n=2). Ten patients were invited to  
 13 participate in the interviews, of whom four declined due to practical reasons. Thus, six  
 14 patients participated in the individual interviews (of which four patients completed the  
 15 programme and two dropped out). In addition, two partners participated in the individual  
 16 interviews. Of the other 19 patients who were asked to participate in the focus groups, seven  
 17 patients declined to participate for practical reasons. Thus, 12 patients participated in the  
 18 focus group interviews (see Table 1 for details). Taken together, 18 out of 29 patients took  
 19 part in the interviews (individual or focus group).

20 Table 1: Participants in the qualitative interviews.

Participants	Patient (individual interviews) Sex (F/M) Age category diagnoses, work	Patients (focus group) Sex, age category, diagnoses, work	Partners (individual interviews) Sex, age category, work	Healthcare professionals
Rehabilitation center	1. F, 70-80 years, FSHD, not working	1. F, 40-50 years, FSHD, not working	1. M, 70-80 years not working	Individual interviews: Rehabilitation physician (n=1) Occupational therapist (n=1)

<b>Klimmendaal, Arnhem</b>	2. F, 60-70 years, IBM, not working	2. M, 60-70 years, HMSN, working 3. M, 60-70 years, MM, working		Physical therapist (n=1)
<b>Community health center Buitenlust, Venray</b>	3. F, 60-70 years CPEO, not working, drop out from intervention group	4. F, 60-70 years, MM, not working 5. F, 30-40 years, MM, working 6. M, 60-70 years, HMSN, not working 7. M, 40-50 years, FSHD, Working	-	Individual interviews: Occupational therapist (n=1) Physical therapist (n=1)
<b>Radboud University Medical Center, Nijmegen</b>	4. M, 50-60 years, MM, not working 5. F, 60-70 years, IBM, not working 6. F, 50-60 years, myasthenia gravis, working 7. M, 30-40 years, MM, not working dropped out from intervention group	8. M, 60-70 years, FSHD, working 9. M, 40-50 years, MD, not working 10. F, 30-40 years, MD, working 11. F, 70-80 years, FSHD, working 12. F, 40-50 years, HMSN, not working	2. F, 60-70 years, working	Individual interviews: Rehabilitation physician (n=1) Occupational therapists (n=2) Neurologist (n=1) Internist (n=1) Secretary (n=1) Physical therapist (n=1) Member patient support association (n=1)
<b>Total interviews</b>	N =7	N= 12	N=2	N= 13

1 M= male, F= female, FSHD = facioscapulohumeral dystrophy; IBM = inclusion body myositis; MM =  
2 mitochondrial myopathy; MD= myotonic dystrophy type 1; CPEO = chronic progressive external  
3 ophthalmoplegia; HMSN = hereditary sensory motor neuropathy

#### 5 *Healthcare professionals*

6 All thirteen healthcare professionals involved in the recruitment, organisation and delivery of  
7 the Energetic programme were interviewed. One professional was involved in the logistics  
8 and planning (secretary), five professionals were involved in the recruitment (four physicians  
9 and one representative of the patient association), and seven professionals were involved in  
10 the delivery of the programme (three PT and four OT).



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## 2 **Satisfaction questionnaire**

3 The analysis of the satisfaction questionnaire (Table 2) showed that 96% of the patients were  
4 entirely or largely satisfied with the results of the intervention. The mean grade of satisfaction  
5 with Energetic was 8.7 (SD 1.1) (scale 1-10). Management of the impairments was perceived  
6 as “entirely” or “largely” improved by 88% of the patients and the Energetic programme was  
7 “largely” (32%) or “entirely” (68%) recommended to others/peers. Regarding the content of  
8 Energetic, patients were overall satisfied with the number and length of the sessions, as well  
9 as with the therapists and the different modules. In total, 24% of the patients evaluated the  
10 total period of 16 weeks as too short, whereas 20% considered the number of sessions in the  
11 implementation and relapse prevention module as too high. The sessions on nutrition and  
12 work were rated lower than other sessions.

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1 Table 2: Patient satisfaction questionnaire regarding the results, delivery and content of the  
 2 Energetic programme.

Satisfaction with the results †	Respondents N	Entirely	Largely	Slightly	Not at all
Satisfaction with the intervention results	25	18 (73%)	6 (25%)	1 (2%)	0
Better management of impairments	25	13 (52%)	9 (36%)	3 (12%)	0
Recommendation of Energetic	25	17 (68%)	8 (32%)	0	0
<b>Satisfaction with the number of sessions†</b>		<b>Just right</b>	<b>Too few/too short</b>	<b>Too many/too long</b>	
Per week	25	25 (100%)	0	0	
Aerobic exercise training	25	23 (92%)	1 (4%)	1 (4%)	
Physical education	25	20 (80%)	2 (8%)	3 (12%)	
Energy conservation management	24	21 (84%)	1 (4%)	2 (8%)	
Implementation relapse prevention	25	17 (68%)	3 (12%)	5 (20%)	
Total period (16 weeks)	25	19 (76%)	6 (24%)	0	
<b>Satisfaction with the length of the sessions†</b>					
Fatigue management	24	16 (64%)	6 (24%)	2 (8%)	
Aerobic exercise training	24	18 (72%)	4 (16%)	2 (8%)	
Education about aerobic exercise	24	20 (80%)	2 (8%)	2 (8%)	
<b>Involvement of partner/next of kin in Energetic †</b>	23	21 (84%)	2 (8%)	0	
<b>Rating the content of the sessions ‡</b>		<b>Mean rate 1-10 (SD)</b>			
Energy conservation management	24	7.6 (1.7)			
Aerobic exercise training	24	8.8 (1.1)			
Education on aerobic exercise	24	8.4 (1.0)			
Experience regular sports	25	7.9 (1.4)			
Food and nutrition	25	6.3 (2.4)			
Work/employment	22	6.1(2.5)			
Location/facilities	25	8.4 (1.2)			
Physical therapists: physical training	24	8.9 (1.0)			
Occupational therapist: Energy conservation management	24	8.7 (1.2)			
<b>Total programme Energetic</b>	<b>25</b>	<b>8.7 (1.1)</b>			

3 † Percentage responding patients with the number and length of sessions. ‡Mean rating  
 4 (standard deviation) of the content of the sessions by patients.

## 6 Interviews

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2  
3 1 The interviews with patients, partners and healthcare professionals resulted in five main  
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5 2 themes. Table 3 shows an overview of the main themes, subthemes and quotes. Each theme is  
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7 3 described and supported by quotes from the participants.  
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For peer review only

1 Table 3: Overview of patients', partners' and healthcare professionals' perspectives regarding  
 2 the content and delivery of the Energetic programme

Themes and subthemes	
1.	<p>The combination of modules makes a complete picture.</p> <p>A. The combination of physical training and fatigue management gives insight into one's capacities  <i>"The combination creates a complete picture," (PAN1).</i>  <i>"I could, therefore, still participate in the cake decoration course, if I plan it in as part of my day programme." (PAA5).</i>  <i>"After illness I have to slowly build the physical training programme up to a given point." (PAA5).</i>  <i>"He has learnt to cope better mentally and that was very important for me." (CG2).</i></p> <p>B. Being prepared to change lifestyle is pivotal  <i>"We were all prepared to change things in our lifestyle, to adapt things and to try new things," (PAV2).</i></p> <p>C. Sustainability of implementation in daily life is essential  <i>"A step-by-step guide was provided that could easily be applied practically," (PAN8).</i>  <i>"Good to correct the entrenched deviations and also ask a huge amount of questions," (PAA5).</i></p> <p>D. Sport's participation in one's own environment challenging  <i>"They had already approached clubs before we had finished the Energetic programme," (OT).</i>  <i>"The different sports activities could be placed earlier in the programme, so that I can find out what is good for me." (PAN2).</i></p>
2.	<p>The programme is physically and mentally intensive  <i>"The conversation was exhausting, more difficult and more confrontational than expected," (PAA3).</i>  <i>"The afternoon hours with the occupational therapist were often a bit tedious" (PAA5)</i>  <i>"The session on nutrition could have been more" (PAN2)</i>  <i>"I had underestimated the time. It cost me a lot more time than expected. This made it difficult to plan in with my home situation." (PAV5).</i>  <i>"All consultations were useful and important for me" (PAA3).</i>  <i>"The Energetic programme is an extremely complex programme to plan and organise" (SC).</i></p>
3.	<p>The group setting is valuable  <i>"We learn from each other" (PAV5),</i>  <i>"We encourage each other" (PAA5)</i>  <i>"Within the group, different approaches and viewpoints are heard – it does not always come from the therapist" (OT).</i>  <i>"In future, there needs to be more time for individual questions of all participants, because this time there were two people who dominated time with their questions about looking for work and hobby participation," (OT).</i></p>
4.	<p>Small variation in delivery in different settings  <i>"Some of the bicycles are a little heavier, then patients (from other therapists) just had to go on those because my patients need a lighter bike. This was always discussed and ended up not being a problem," (PT).</i>  <i>"I found that planning the sports sessions was always labour intensive and needed a lot of explanation towards management," (PT).</i></p>

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3 1 5. Therapists are coaches  
4 "allowing us to think outside of the box" (PAA5).  
5 2 "real interest" (PAV2),  
6 "expertise" (PAA4),  
7 3 "Guiding a group and encouraging 'change language' is not something you can  
8 4 learn really quickly and easily in one training session," (OT)  
9 5 A. Therapists need education  
10 "It was really stimulating in terms of the learning activities; it was varied in theory  
11 6 and practice in terms of what needed to be done on the programme," (PT).  
12 7 "You obtain the information, but you can only really engage in conversation about it  
13 when you have tried to apply it yourself," (PT).  
14 "Then you can hear from everyone about how it went," (OT).  
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18 8 Patients (PA), partners or next of kin (NoK) and healthcare professionals, including occupational therapists  
19 9 (OT), physical therapists (PT), physicians (PHYS), and secretary (SC), were given an individual number to  
20 10 ensure their anonymity. The setting was indicated as Nijmegen (N), Arnhem (A), or Venray (V).  
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## 23 11 Themes

### 24 12 1. The combination of modules makes a complete picture.

#### 25 13 1.A. The combination of physical training and fatigue management gives insight into 26 14 one's capacities

27 15 An important characteristic of Energetic for both patients and therapists was the combination  
28 16 of the four modules. Improvement of physical fitness, education about AET, applying ECM  
29 17 strategies, and implementation of advice, training and strategies in daily life helped patients to  
30 18 get insight in their energy levels and physical capacities.  
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33 19 "The combination creates a complete picture." (PANI).

34 20 Patients reported that their participation level had increased, because they had become more  
35 21 aware of their possibilities to manage their energy in daily life.  
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38 22 "I could, therefore, still participate in the cake decoration course, if I plan it in as part  
39 23 of my day programme." (PAA5).  
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1 The majority of patients reported that their physical fitness had improved. Additionally, most  
2 patients had become more aware of their own physical limitations and had gained a better  
3 understanding of how they could manage these limitations during physical activities.

4 *“After illness I have to slowly build the physical training programme up to a given  
5 point.” (PAA5).*

6 The two partners reported that their spouses had learned to better cope with the symptoms.

7 *“He has learnt to cope better mentally and that was very important for me.” (NoK2).*

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9 *I.B. Being prepared to change lifestyle is pivotal*

10 Before the start of the programme both patients and therapists committed themselves to  
11 participation. An intake assessment was held with the patients to determine if they would be  
12 able to participate in the programme for 16 weeks and willing to integrate what they would  
13 learn into their everyday lives. They also identified what they wanted to achieve through  
14 participation in the Energetic programme.

15 *“We were all prepared to change things in our lifestyle, to adapt things and to try new  
16 things.” (PAV2).*

17  
18 *I.C Sustainability of implementation in daily life is essential*

19 The steady structure in the programme, the translation to practical situations and the  
20 integration in daily routines were experienced as valuable.

21 *“A step-by-step guide was provided that could easily be applied practically.” (PAN8).*

22 Three months after completion of the programme there was a booster session, which was  
23 valued as positive by most patients.

1  
2  
3 1 “Good to correct the entrenched deviations [after three months] and also ask a huge  
4  
5 2 amount of questions.” (PAA5).  
6  
7

8 3 Several patients reported that they would appreciate more booster sessions in the future to be  
9  
10 4 better able to retain the newly learnt behaviours in everyday life.  
11  
12

13 5  
14  
15  
16 6 *1.D. Sports participation in one’s own environment is challenging*

17  
18 7 An important element of the Energetic programme is the guidance offered to implement  
19  
20 8 sports activities in everyday life. Therapists reported that most patients actively sought  
21  
22 9 possibilities for sports participation. Some patients actually joined a sport in their own  
23  
24 10 environment after completing the programme.  
25  
26

27  
28 11 “They had already approached clubs before we had finished the Energetic  
29  
30 12 programme.” (OT).  
31  
32

33 13 However, others reported that they could not find a suitable sports activity. They expressed  
34  
35 14 the wish for more support in seeking appropriate sports activities in their neighbourhood.  
36  
37

38 15 The sports sessions that were presented as part of the programme were positively valued, but  
39  
40 16 also difficult to perform. A few patients would have liked the sports sessions to be presented  
41  
42 17 earlier in the programme, so that they would have had more time to search and implement an  
43  
44 18 appropriate sport.  
45  
46  
47

48 19 “The different sports activities could be placed earlier in the programme, so that I can  
49  
50 20 find out what is good for me.” (PAN2).  
51  
52

53 21

54  
55  
56 22 2. The programme is physically and mentally intensive  
57  
58  
59  
60

1  
2  
3 1 Both patients and healthcare professionals described Energetic as an intensive programme on  
4  
5 2 many levels. Most patients mentioned the physical training as a factor that contributed to the  
6  
7 3 intervention burden. In addition, the mental strain of having to evaluate and reflect on one's  
8  
9 4 own behavioural patterns was also experienced as burdensome.

10  
11  
12  
13 5 *“The conversation was exhausting, more difficult and more confrontational than*  
14  
15 6 *expected.” (PAA3).*

16  
17  
18 7 Some patients reported that the frequency and duration of the sessions were exhausting in the  
19  
20 8 context of everyday life, whereas others reported that Energetic fitted well within their daily  
21  
22 9 routine. By some, the travel distance to the programme was mentioned as a stressor.

23  
24  
25 10 *“I had underestimated the time. It costed me a lot more time than expected. This made*  
26  
27 11 *it difficult to plan in with my home situation.” (PAV5).*

28  
29  
30 12 Regarding the content of the programme, some patients mentioned that the ECM sessions  
31  
32 13 were long and contained repetitions of theories.

33  
34  
35 14 *“The afternoon hours with the occupational therapist were often a bit tedious”*

36  
37  
38 15 For some patients, the session on the topic of work was not applicable as they were no longer  
39  
40 16 working (after retirement or cessation of work due to the consequences of NMD). The session  
41  
42 17 on nutrition was perceived by some patients as too short and, therefore, lacking depth.

43  
44  
45 18 *“The session on nutrition could have been more”*

46  
47  
48 19 Nevertheless, most patients reported that they would not like to see any element of the  
49  
50 20 programme being deleted.

51  
52  
53 21 *“All consultations were useful and important for me.” (PAA3).*

54  
55  
56 22 The healthcare professionals reported the complex planning of the programme within their  
57  
58 23 work schedule as intensive.



1  
2  
3 1 *“The Energetic programme is an extremely complex programme to plan and*  
4  
5 2 *organise” (SC).*  
6  
7  
8 3  
9

10  
11 4 3. *The group setting is valuable*  
12

13 5 All patients and healthcare professionals reported that they experienced the group setting as  
14  
15 6 valuable in order to share experiences, to learn from others, and to motivate each other.  
16  
17

18 7 *“We learn from each other” (PAV5), “We encourage each other” (PAA5) “Within the*  
19  
20 8 *group, different approaches and viewpoints are heard – it does not always come from*  
21  
22 9 *the therapist.” (OT).*  
23  
24  
25

26 10 Both therapists and patients reported a group of six patients as optimal to be able to focus on  
27  
28 11 all personal dilemmas and questions. One patient and one therapist explicitly expressed that  
29  
30 12 they did not have enough time for individual questions.  
31  
32

33 13 *“In future, there needs to be more time for individual questions of all participants,*  
34  
35 14 *because this time there were two people who dominated time with their questions*  
36  
37 15 *about looking for work and hobby participation.” (OT).*  
38  
39  
40  
41 16

42  
43 17 4. *Small variations in delivery occur in different settings*  
44

45 18 The Energetic programme was offered in a variety of clinical settings. Patients and therapists  
46  
47 19 reported that practical solutions needed to be found at the various locations.  
48  
49

50  
51 20 *“Some of the bicycles are a little heavier. Then patients (from other therapists) just*  
52  
53 21 *had to go on those because my patients needed a lighter bike. This was always*  
54  
55 22 *discussed and ended up not being a problem.” (PT).*  
56  
57  
58  
59  
60

1  
2  
3 1 It also became apparent that the different clinical settings organised certain details differently.  
4  
5 2 At the rehabilitations centre, for example, five minutes of Tai Chi was performed by means of  
6  
7 3 a warm up before the training. The sports sessions were organised at a regular sports complex  
8  
9 4 outside the centre. The costs hereof were not covered by the medical insurance and therapists  
10  
11 5 from two settings experienced this as a draw-back.  
12  
13

14  
15 6 *“I found that planning the sports sessions was always labour intensive and needed a*  
16  
17 7 *lot of explanation towards management.” (PT).*  
18  
19

## 20 8

### 21 9 5. Therapists are coaches

22  
23 10 Patients and therapists alike reported that therapists adopted the role of a ‘coach’ during the  
24  
25 11 programme. The therapists also reported that the collaboration between them (OT and PT)  
26  
27 12 was important to guide the group well.  
28  
29

30  
31 13 The therapist’s characteristics that patients found important were “*expertise*” (PAA4), “*real*  
32  
33 14 *interest*” (PAV2), and “*allowing us to think outside of the box*” (PAA5). A few patients  
34  
35 15 reported that they found the guidance from the therapists insufficient and lacking attention  
36  
37 16 regarding individual differences. Additionally, some therapists reported that supervising an  
38  
39 17 entire group required a lot of attention.  
40  
41  
42

43  
44 18 *“Guiding a group and encouraging ‘change language’ is not something you can learn*  
45  
46 19 *really quickly and easily in one training session.” (OT).*  
47  
48

#### 49 20

#### 50 21 5.A. *Therapists need education*

51  
52 22 The education programme for therapists was experienced as valuable.  
53  
54

55  
56 23 *“It was really stimulating in terms of the learning activities; it was varied in theory*  
57  
58 24 *and practice in terms of what needed to be done on the programme,” (PT).*  
59  
60

1 A few therapists reported that they would have preferred less time between workshop sessions  
2 to prepare for the Energetic programme in their own setting. They liked obtaining the theory  
3 and then being able to implement in daily life what they had learnt.

4 *“You obtain the information, but you can only really engage in conversation about it  
5 when you have tried to apply it yourself.” (PT).*

6 The therapists reported that interaction among peers was important for everyone’s learning  
7 process. The practice experiences were shared during the group supervision.

8 *“Then you can hear from everyone about how it went’.” (OT).*

## 10 DISCUSSION

11 A mixed methods evaluation of the Energetic programme showed a diverse picture of the  
12 facilitators and barriers related to the content and delivery of this multidisciplinary out-patient  
13 group intervention for patients with NMD and chronic fatigue.

14 The patients’ insight in their own capacities and improved participation level was consistent  
15 with the aim of the Energetic programme and with the observed improvement on the primary  
16 outcome of our RCT, the COPM.<sup>6</sup> The COPM measures experienced problems in activities  
17 that are important and meaningful for an individual.<sup>19 20</sup> The choice of the COPM as a primary  
18 outcome fits with the client-centred approach of Energetic and with the impact reported by  
19 patients in this evaluation. Moreover, the perceived improvement of physical fitness reported  
20 by patients was in line with the observed improvement of physical endurance as measured  
21 with the six-minute walking test in our RCT.<sup>6</sup> To measure insight into patient’s own  
22 capacities, the general self-efficacy scale (GSES) was used, which showed no group  
23 difference or change over time. However, the GSES is not specifically designed for the self-  
24 efficacy to implement energy conservation strategies. An alternative self-efficacy assessment

1 developed by Liepold et al.<sup>21</sup> specifically evaluates self-efficacy in performing energy  
2 conservation management strategies and might be a possible valuable measure in future  
3 programme evaluations.

4 Patients and healthcare professionals reported that the group setting supported the patients to  
5 learn from their own experiences, as well as from each other, with the therapists taking the  
6 role of a coach. Such vicarious experiences, including verbal (social) persuasion, fit well with  
7 Bandura's self-efficacy theory,<sup>22</sup> and are believed to support behavioural change. In addition,  
8 guidance in embedding AET and ECM strategies by self-monitoring behaviour and receiving  
9 feedback from peers may contribute to patients' self-management capacities<sup>9 23</sup>. However, in  
10 this evaluation, some patients reported that, despite this guidance, they found it difficult to  
11 implement exercising at home and maintain the acquired skills in the long term. This is in line  
12 with a study of Wallace where patients mentioned a high motivation to maintain exercising  
13 after a training programme, but experienced barriers to gym membership and  
14 implementation<sup>24</sup>. This phenomenon has been described by Packer, who emphasised that self-  
15 management is an ongoing process requiring continuous effort and support to gain  
16 knowledge, skills and confidence over time.<sup>25</sup> Additional booster sessions are, therefore,  
17 recommended to enable trial-and-error practice in a constantly changing context and to  
18 receive encouragement from peers and knowledgeable healthcare professionals. These booster  
19 sessions should focus on the maintenance of exercising, planning, and pacing in daily life  
20 taking into account the progressive character of the disease and the changing roles and  
21 context.

22 During the interviews, patients reported a high willingness to change before the start of the  
23 programme, which was probably related to the motivational screening by occupational  
24 therapists before participation. The screening for (in)eligibility before the start of the RCT  
25 regarded the individual motivation to change behaviour and the expected individual

1 intervention burden, which resulted in an exclusion of 43 patients.<sup>6</sup> Nevertheless, in the  
2 interviews, the Energetic programme was reported to be physically and mentally intensive  
3 and sometimes difficult to schedule within the weekly agenda, which also depended on travel  
4 distance. This perceived intervention burden is an important factor for patients' willingness to  
5 participate in Energetic and should be clear during the screening for patients at the start of the  
6 programme. However, in the interviews, patients reported that no elements should be taken  
7 out of the programme. A way to reduce the intervention burden would be the use of blended  
8 care, for instance combining e-health and face-to-face sessions.<sup>26 27</sup> The recent developments  
9 during the COVID-19 pandemic, for instance the increase in video calls for regular  
10 healthcare, show that e-health can be used in combination with traditional forms of care in  
11 outpatient rehabilitation.<sup>28</sup>

12 The results of this study suggest that Energetic can be delivered in a rehabilitation centre, in a  
13 specialised hospital department, as well as in a primary care setting. Only minor practical  
14 adjustments were necessary per setting. The collaboration among therapists within and  
15 between settings was considered to be a facilitating factor for the delivery of the programme.  
16 This is in line with the study by van Dongen et al.,<sup>29</sup> who identified facilitating factors for  
17 interdisciplinary collaboration, such as knowing each other well, organisational factors  
18 regarding the delivery of a intervention and professional meetings, and having a shared  
19 vision. Additionally, they stressed the importance of a team leader who plays a key role in  
20 overseeing the organisation and guiding the team through the developments.<sup>29</sup> Due to the  
21 complexity in the organisation and planning of the programme we, therefore, suggest that  
22 every clinical setting should assign a team leader to implement Energetic.

23 Both therapists and patients experienced that guiding a group requires specific skills for  
24 therapists. Therapist are trained in individual consultations with patients, suggesting that  
25 specific group didactic skills would be helpful to optimize the group interaction within

1  
2  
3 1 Energetic Finally, finding finances for the external sports sessions was reported by therapists  
4  
5 2 as a barrier. Regular sports activities in society are organised outside the healthcare setting  
6  
7 3 and, thus, are not within the traditional scope of most therapists and not financially  
8  
9  
10 4 reimbursed by healthcare insurances. For better implementation of this aspect of the Energetic  
11  
12 5 programme, collaboration with governmental sports organisations and healthcare  
13  
14 6 professionals working in regular sports domains should be considered.<sup>30</sup>  
15  
16  
17 7

## 8 Strengths and limitations

9 We tried to optimise the credibility of our results by including the perspectives of patients,  
10 partners, and healthcare professionals.<sup>31</sup> Furthermore, we used independent interviewers for  
11 the individual and focus group sessions, independent research assistants to establish the  
12 coding structure's validity, and we emailed the themes to all participants and asked if they  
13 could identify themselves with these themes. We have followed different strategies to  
14 enhance the trustworthiness of the findings: triangulation of data collection methods and  
15 triangulation of researchers (use of two researchers for data collection and analysis).  
16 Furthermore, reflective meetings with the research group to discuss the analytical process and  
17 the preliminary and final themes enhanced the credibility of our data.<sup>32 33</sup> Nevertheless,  
18 qualitative research and satisfaction questionnaires reflect the perceived impact and  
19 interpretation of the programme by patients and professionals, which does not allow causal  
20 inferences. Another methodological limitation is that only two partners participated, which  
21 inevitably has led to lack of saturation regarding the partners' perspectives. In addition, the  
22 fact that the satisfaction questionnaires were only filled in by patients who completed the  
23 intervention can be considered a methodological limitation, because it may have led to  
24 selection bias. Yet, we gained some insight also in the experiences of those who dropped out

1 by interviewing two patients that discontinued the intervention because of its intensity or due  
2 to co-morbidities.

3

#### 4 **CONCLUSION**

5 This mixed methods evaluation was conducted to investigate the experiences of patients and  
6 healthcare professionals involved in a multidisciplinary out-patient self-management group  
7 programme called Energetic. The aim of this programme is to improve social participation  
8 and physical endurance in people with NMD and chronic fatigue. Patients were overall  
9 satisfied with the number and length of the sessions, as well as with the therapists and the  
10 different modules. Our results indicate that Energetic can be implemented in different clinical  
11 settings and that the use of group sessions and using a combination of AET, education about  
12 AET, ECM, and daily-life implementation are facilitators for attaining better self-  
13 management. Patient suggestions for programme improvement are the use of blended care  
14 interventions, inclusion of more booster sessions, and more guidance in seeking appropriate  
15 sports activities in the personal environment. As for the therapists, suggested improvements  
16 included enhancement of group supervising skills and collaboration between therapists and  
17 society or governmental sports organisations.

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## 14 **Availability of data and material**

15 All data used and analysed during the current study are available from the corresponding  
16 author on reasonable request until 2026.

## 17 **Declaration of Conflicting Interests**

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### 37 38 39 40 17 **Patients and public partnership**

41  
42 18 The Energetic programme is developed for and with patients in the outpatient’s rehabilitation  
43  
44 19 clinic of the department rehabilitation Radboudumc. Patients were not involved in  
45  
46 20 development of this mixed method evaluation on the Energetic programme. However, the  
47  
48 21 experiences of patients and health professionals were carefully selected in this study to  
49  
50 22 improve the intervention and further implementation.

### 51 52 53 54 24 **Authors’ contributions**

1 YV, EHCC, BGMvE and ACHG conceptualised the study and coordinated funding. YV  
2 undertook and monitored the study with supervision from EHCC, JTG, ACHG, TS and  
3 MNvS. YV was responsible for supervising the therapists, research assistants, and recruited  
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5 YV and EHCC, TS and MNvS wrote the first draft of the manuscript and were responsible for  
6 revisions. YV, EHCC, TS, MJLG, MNvS, BGMvE and ACHG discussed and commented on  
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3 **1 Tables/figures**

4  
5 2 Table 2: Participants in the qualitative interviews.

6  
7 3 FSHD = facioscapulohumeral dystrophy; IBM = inclusion body myositis; MM mitochondrial  
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9 4 myopathy; MD= myotonic dystrophy type 1

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12 6 Table 2: Patient satisfaction questionnaire regarding the perceived delivery and content of the  
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14 7 Energetic programme.

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16 8 † Percentage responding patients with the number and length of sessions. ‡Mean rating  
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18 9 (standard deviation) of the content of the sessions by patients.

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22 11 Table 3: Overview of patients', partners' and healthcare professionals' perspectives regarding  
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24 12 the content and delivery of the Energetic programme

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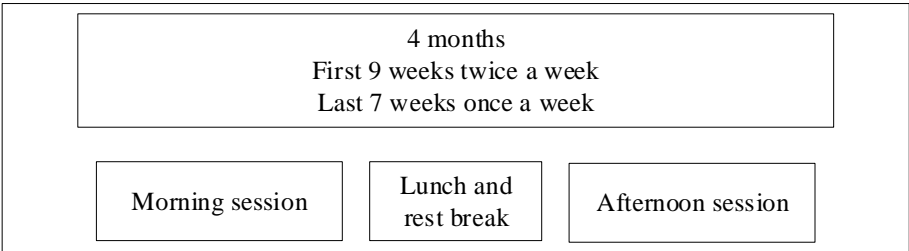
27  
28 14 Figure 1: Content of the Energetic programme delivered by physical therapists and occupational  
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30 15 therapists.

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**Inclusion criteria for the Energetic programme:**

- Patients with a NMD and chronic fatigue influencing their social participation
- Motivation and readiness to change explored by motivational interviewing
- No depressive symptoms or other psychiatric or cognitive symptoms as judged by a psychologist
- Formulation of at least three personalized goals
- Physical capacity to participate in aerobic exercise training based on a cardiopulmonary test.



<p><b>Aerobic exercise training</b> 9 weeks; 2 times weekly (18 sessions) 7 weeks; once a week (7 sessions) Duration: 90 minutes Trainer: Physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Individually tailored exercise training at 50-70% of the maximum heart rate, guided by a cardiac rhythm monitor</li> <li style="padding-left: 20px;">- Fine tuning takes place based on the recovery rate</li> <li>- Different exercises depending on the reference and motor abilities of the patients.</li> </ul>	<p><b>Energy conservation management[5]</b> Weeks 2-8 and 10 (8 sessions) Duration; 90 minutes Trainer: Occupational therapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>-Benefits of rest</li> <li>- Effectively Communicating with the social environment</li> <li>- Applying principles of proper body mechanics and ergonomics</li> <li>- Adequately modifying the personal environment</li> <li>- Analysing and adjusting individual activities</li> <li>- Setting priorities</li> <li>-Finding an activity-rest balance over the entire day and week</li> <li>- Setting short-term and long-term goals and action planning</li> </ul>
<p><b>Education about aerobic exercise</b> 3 weeks; once a week (3 sessions) Duration: 60 minutes Trainer: Physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Attaining an adequate training stimulus</li> <li style="padding-left: 20px;">- The need to rest and recuperate</li> <li>- Designing and adhering to a feasible training programme</li> <li style="padding-left: 20px;">- Prevention of overtraining and relapse</li> <li>- Management physical training after relapse</li> </ul>	<p><b>Relapse prevention and implementation and in daily life</b> Weeks 5-13 and 16 (8 sessions) Duration training: 60 minutes Trainer: Occupational therapist and physiotherapist</p> <p>Content of the sessions:</p> <ul style="list-style-type: none"> <li>- Education by a dietician</li> <li>- NMD and fatigue regarding work</li> <li>- Two sessions with the partners/next of kin</li> <li>- Different types of exercise to explore (swimming, Nordic walking, yoga, body work-out, dancing)</li> <li>- Explore possibilities for exercising in patients own environment</li> </ul>

**A: Satisfaction questionnaire****Planning and organization**

Question	Answer options
Was the total number of days per week adequate for you?	Just right Too few/too short Too many/too long
Was the total number of physical training sessions adequate for you?	Just right Too few/too short Too many/too long
Was the total number of physical education meetings adequate for you?	Just right Too few/too short Too many/too long
Was the total number of sessions for energy conservation management adequate for you?	Just right Too few/too short Too many/too long
Was the total number of sessions for implementation and relapse prevention adequate for you?	Just right Too few/too short Too many/too long
What did you think of the length of the period (16 weeks) in which the treatments took place?	Just right Too few/too short Too many/too long
How do you rate the location and facilities?	Rate 1= extremely bad; 10= extremely good

**Module Energy conservation management**

Question	Answer options
How do you rate session 1: Importance of rest?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 2: Communication, postures and positioning?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 3: Practical situations?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 4: Priorities/standards/norms and values and analysis/adaptation of activities?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 5: Balance in your schedule?	Rate 1= extremely useless; 10= extremely useful
How do you rate session 6: Evaluation and future plans?	Rate 1= extremely useless; 10= extremely useful
How do you rate the length of session's energy conservation management?	Just right Too few/too short Too many/too long
How do you rate the way the sessions were supervised by the occupational therapist?	Rate 1= extremely useless; 10= extremely useful

**Module Aerobic exercise training**

Question	Answer options
How do you rate the added value/use of the physical training?	Rate 1= extremely useless; 10= extremely useful
How do you rate the way in which the sessions were supervised by the physical therapist?	Rate 1= extremely useless; 10= extremely useful
What did you think of the length of the physical training sessions?	Just right Too few/too short Too many/too long

**Module physical education**

Question	Answer options
How do you rate session 1: Introduction and training theory	Rate 1= extremely useless; 10= extremely useful
How do you rate session 2: Effects of training	Rate 1= extremely useless; 10= extremely useful



How do you rate session 3: Completion of the training & preparation of a training schedule	Rate 1= extremely useless; 10= extremely useful
What did you think of the length of the physical education sessions?	Just right Too few/too short Too many/too long
How do you rate the way in which the sessions were supervised by the physical therapist?	Rate 1= extremely useless; 10= extremely useful

### Module implementation and relapse prevention

Question	Answer options
What did you think of the extent to which your next of kin or partner was involved in the programme?	Just right Too few/too short Too many/too long
How do you rate the various sports sessions?	Rate 1= extremely useless; 10= extremely useful
How do you rate the dietetics/nutrition session?	Rate 1= extremely useless; 10= extremely useful
How do you rate employment session?	Rate 1= extremely useless; 10= extremely useful

### Therapists

Question	Answer options
Did you find the occupational therapist competent?	No, not at all A little Largely so Yes, entirely
Did the occupational therapist give advice that is appropriate and useful for your situation?	No, not at all A little Largely so Yes, entirely
Did you find the physical therapist competent?	No, not at all A little Largely so Yes, entirely
Did the physical therapist give advice that is suitable and useful for your situation?	No, not at all A little Largely so Yes, entirely

### Treatment

Question	Answer options
Did the occupational therapist treat you politely and with respect?	No, not at all A little Largely so Yes, entirely
Did the occupational therapist make you feel at ease?	No, not at all A little Largely so Yes, entirely
Did the physical therapist treat you politely and with respect?	No, not at all A little Largely so Yes, entirely
Did the physical therapist ensure that you felt at ease?	No, not at all A little Largely so Yes, entirely

### Results

Question	Answer options
Are you satisfied with the results of the Energetic programme?	No, not at all A little Largely so Yes, entirely
Can the Energetic programme help you deal with your limitations and/or problems better than before?	No, not at all A little Largely so Yes, entirely

**Overall satisfaction**

Question	Answer options
Suppose you have a good friend who is in the same situation as you. Would you recommend this friend to participate in the Energetic programme?	Yes, absolutely Yes, maybe No I don't know if I would do that
How do you rate your satisfaction with the Energetic programme?	Rate 1= extremely bad; 10= extremely good

**Open questions:**

- The most valuable for me was:
- If I could change the programme, I would change....
- Space for comments on the Energetic programme

**Appendix B: Interview guides.**

Individual interviews with patients and partners	
Perceived impact	What is your experience with Energetic?
Content of Energetic	What were the positive and negative aspects of the content of Energetic? Elements: Experience with Energetic, different modules, frequency, lengths, construction, group, goal setting, next of kin's involvement, influence on home situation, trainers
Organisation	What were the facilitators and barriers in the delivery and organisation of Energetic? Elements: location, accessibility, travel time, facilities,
Focus group with patients	
	<p>How did you experience the quality of:</p> <ul style="list-style-type: none"> <li>• Content of the sessions</li> <li>• Working methods</li> <li>• Education</li> <li>• Individual goals</li> <li>• Group setting</li> <li>• Trainers</li> <li>• Organization</li> <li>• involvement of next of kin/partners</li> </ul> <p>Creative questions regarding the benefit of Energetic. For instance:</p> <ul style="list-style-type: none"> <li>• If you could make a commercial about Energetic for the health insurance, what would you mention in this commercial?</li> <li>• Suppose you are in the waiting room in the hospital and would meet a peer, what would you tell him/her about Energetic?</li> <li>• Suppose in one-month time you are back with the rehabilitation physician who referred you to Energetic, what will you tell him/her about the programme?</li> <li>• What components should stay in Energetic?</li> </ul> <p>Barriers:</p> <ul style="list-style-type: none"> <li>• If the health insurance company forces you to adapt the programme in order to reduce costs, what components could be deleted from Energetic?</li> <li>• What components could be altered or deleted from Energetic?</li> </ul> <p>Addition:</p> <ul style="list-style-type: none"> <li>• For instance: if money is no issue for Energetic, what would you add to the programme?</li> <li>• Is there some component that you missed in Energetic?</li> <li>• If you must describe your experience with Energetic in a few words, what would that be?</li> </ul>
Individual interviews with healthcare professionals	

All healthcare professionals	General information: name, function in relation to Energetic.
Healthcare professionals involved in the delivery	<p>How was the starting phase (implementation) of the programme?</p> <p>What were the facilitators and barriers for Energetic at your location?</p> <p>What is needed in the programme to implement it nationally?</p> <p>Elements: provided means for implementation, instruction manual, education, meetings, time investment, what should be changed or altered in the starting phase of Energetic (implementation), guidance as a therapist, communication in the programme (in the setting and between the different settings)</p>
Healthcare professionals involved in the recruitment	<p>Can you describe how the recruitment took place?</p> <p>What is needed to recruit patients for Energetic?</p> <p>Can you tell us something about the time investment for recruitment?</p>
Healthcare professionals involved in the organisation	<p>Can you describe how the organisation took place?</p> <p>What means and facilities were necessary to organise Energetic and how did this influence the delivery of the programme?</p>

**The TIDieR (Template for Intervention Description and Replication) Checklist\*:**

Information to include when describing an intervention and the location of the information

Item number	Item	Where located **	
		Primary paper (Page or appendix number)	Other † (details)
1.	<b>BRIEF NAME</b> Provide the name or a phrase that describes the intervention.	Page 2, 5, 6	_____
2.	<b>WHY</b> Describe any rationale, theory, or goal of the elements essential to the intervention.	Page 4	_____
3.	<b>WHAT</b> Materials: Describe any physical or informational materials used in the intervention, including those provided to participants or used in intervention delivery or in training of intervention providers. Provide information on where the materials can be accessed (e.g. online appendix, URL).	Page 5-7	_____
4.	Procedures: Describe each of the procedures, activities, and/or processes used in the intervention, including any enabling or support activities.	Figure 1, page 5-	_____
5.	<b>WHO PROVIDED</b> For each category of intervention provider (e.g. psychologist, nursing assistant), describe their expertise, background and any specific training given.	Figure 1, page 5-	_____
6.	<b>HOW</b> Describe the modes of delivery (e.g. face-to-face or by some other mechanism, such as internet or telephone) of the intervention and whether it was provided individually or in a group.	Figure 1, page 5-	Veenhuizen Y, Cup EH, Groothuis JT, et al. Effectiveness and cost-effectiveness of a self-management group program to

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<p><b>WHERE</b></p> <p>7. Describe the type(s) of location(s) where the intervention occurred, including any necessary infrastructure or relevant features.</p>	<p>improve social participation in patients with neuromuscular disease and chronic fatigue: protocol of the Energetic study. <i>BMC Neurol</i> 2015; 15: 58. DOI: 10.1186/s12883-015-0314-4.</p> <p>Veenhuizen 2015</p>
<p><b>WHEN and HOW MUCH</b></p> <p>8. Describe the number of times the intervention was delivered and over what period of time including the number of sessions, their schedule, and their duration, intensity or dose.</p> <p><b>TAILORING</b></p> <p>9. If the intervention was planned to be personalised, titrated or adapted, then describe what, why, when, and how.</p> <p><b>MODIFICATIONS</b></p> <p>10.* If the intervention was modified during the course of the study, describe the changes (what, why, when, and how).</p> <p><b>HOW WELL</b></p> <p>11. Planned: If intervention adherence or fidelity was assessed, describe how and by whom, and if any strategies were used to maintain or improve fidelity, describe them.</p> <p>12.* Actual: If intervention adherence or fidelity was assessed, describe the extent to which the intervention was delivered as planned.</p>	<p>Figure 1, Veenhuizen 2015</p> <p>page 20-23 Veenhuizen 2015</p> <p>7/A</p> <p>page 20-23</p> <p>page 20-23</p>

1 \*\* **Authors** - use N/A if an item is not applicable for the intervention being described. **Reviewers** – use ‘?’ if information about the element is not reported/not  
2 sufficiently reported.  
3

4 † If the information is not provided in the primary paper, give details of where this information is available. This may include locations such as a published protocol  
5 or other published papers (provide citation details) or a website (provide the URL).  
6

7 ‡ If completing the TIDieR checklist for a protocol, these items are not relevant to the protocol and cannot be described until the study is complete.

8 \* We strongly recommend using this checklist in conjunction with the TIDieR guide (see *BMJ* 2014;348:g1687) which contains an explanation and elaboration for each item.  
9

10 \* The focus of TIDieR is on reporting details of the intervention elements (and where relevant, comparison elements) of a study. Other elements and methodological features of  
11 studies are covered by other reporting statements and checklists and have not been duplicated as part of the TIDieR checklist. When a **randomised trial** is being reported, the  
12 TIDieR checklist should be used in conjunction with the CONSORT statement (see [www.consort-statement.org](http://www.consort-statement.org)) as an extension of **Item 5 of the CONSORT 2010 Statement**.  
13 When a **clinical trial protocol** is being reported, the TIDieR checklist should be used in conjunction with the SPIRIT statement as an extension of **Item 11 of the SPIRIT 2013**  
14 **Statement** (see [www.spirit-statement.org](http://www.spirit-statement.org)). For alternate study designs, TIDieR can be used in conjunction with the appropriate checklist for that study design (see  
15 [www.equator-network.org](http://www.equator-network.org)).  
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## COREQ (CONsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
<b>Domain 1: Research team and reflexivity</b>			
<i>Personal characteristics</i>			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	
Occupation	3	What was their occupation at the time of the study?	
Gender	4	Was the researcher male or female?	
Experience and training	5	What experience or training did the researcher have?	
<i>Relationship with participants</i>			
Relationship established	6	Was a relationship established prior to study commencement?	
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	
Interviewer characteristics	8	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	
<b>Domain 2: Study design</b>			
<i>Theoretical framework</i>			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	
<i>Participant selection</i>			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	
Sample size	12	How many participants were in the study?	
Non-participation	13	How many people refused to participate or dropped out? Reasons?	
<i>Setting</i>			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	
<i>Data collection</i>			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	
Repeat interviews	18	Were repeat interviews carried out? If yes, how many?	
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	
Field notes	20	Were field notes made during and/or after the interview or focus group?	
Duration	21	What was the duration of the interviews or focus group?	
Data saturation	22	Was data saturation discussed?	
Transcripts returned	23	Were transcripts returned to participants for comment and/or	



Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
<b>Domain 3: analysis and findings</b>			
<i>Data analysis</i>			
Number of data coders	24	How many data coders coded the data?	
Description of the coding tree	25	Did authors provide a description of the coding tree?	
Derivation of themes	26	Were themes identified in advance or derived from the data?	
Software	27	What software, if applicable, was used to manage the data?	
Participant checking	28	Did participants provide feedback on the findings?	
<i>Reporting</i>			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	
Data and findings consistent	30	Was there consistency between the data presented and the findings?	
Clarity of major themes	31	Were major themes clearly presented in the findings?	
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

**Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.**