Optimizing quality of care for patients with ALS and their family caregivers
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Cluster RCT of case management on patients’ quality of life and caregiver strain in ALS

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ABSTRACT

Objectives
To study the effect of case management on quality of life (QOL), caregivers’ strain, and perceived quality of care (QOC) in patients with amyotrophic lateral sclerosis (ALS) and their caregivers.

Methods
We conducted a multicenter cluster randomized controlled trial, with the multidisciplinary ALS care team as the unit of randomization. During 12 months, patients with ALS and their caregivers received case management plus usual care, or usual care alone. Outcome measures were the 40-item ALS Assessment Questionnaire (ALSAQ-40), Emotional Functioning domain (EF); the Caregiver Strain Index (CSI); and the QOC score. These measures were assessed at baseline and at 4, 8, and 12 months.

Results
Case management resulted in no changes in ALSAQ-40 EF, CSI, or QOC from baseline to 12 months. ALSAQ-40 EF scores in both groups were similar at baseline and did not change over time (p = 0.331). CSI scores in both groups increased significantly (p < 0.0001). Patients with ALS from both groups rated their perceived QOC at baseline with a median score of 8, which did not change significantly during follow-up.

Conclusion
Within the context of multidisciplinary ALS care teams, case management appears to confer no benefit for patients with ALS or their caregivers.

Classification of Evidence
This study provides Class III evidence that case management in addition to multidisciplinary ALS care does not significantly improve health-related quality of life of patients with ALS.
INTRODUCTION

There is no cure for amyotrophic lateral sclerosis (ALS); treatment is limited to optimal neuropalliative care, anticipating the relentless progression of muscle weakness and its consequences to daily activities and participation. In the Netherlands, in accordance with international guidelines, most patients with ALS are supported by one of the 43 ALS teams that provide proactive multidisciplinary ALS care. Neuropalliative care is diverse and complex. We know from clinical practice and from QOC evaluations from patients and caregivers that ALS care does not always adequately address the needs of patients and their caregivers.

Case management has been suggested as an innovative strategy to optimize complex care, thereby improving patient satisfaction, caregiver satisfaction, and quality of life (QOL). In case management, an individual supports the patient and caregiver through the complex navigation of available resources to manage the consequences of ALS, with the aim of stabilizing QOL. Case management programs are often directed at patients with serious and life-threatening illness and increasingly incorporate palliative care assessment and interventions.

In this study we investigated the added value of case management to multidisciplinary ALS care. We hypothesized that case management in addition to usual care would improve QOL of patients with ALS, caregivers’ burden, and perceived QOC of patients with ALS and their informal caregivers.

METHODS

Study design
We performed a cluster randomized controlled trial (RCT) with the multidisciplinary ALS team as the unit of randomization (figure 1). Teams as a whole were randomized, instead of individual patients, to avoid readjustment of each team’s treatment procedures during the study period owing to their experiences with case management. The full details of our trial protocol are available upon request from the correspondence address.

The primary research question for this study was to determine whether case management in addition to usual multidisciplinary ALS care would improve the QOL of patients. The secondary research questions were if supplemental case management would affect caregivers’ strain and the QOC of patients and their primary informal caregivers compared with multidisciplinary ALS care alone.

This study design provided Class III evidence for case management in addition to multidisciplinary care on health related QOL for patients with ALS, on caregiver burden and on QOC for patients and their caregivers.
Standard Protocol Approvals, Registrations, and Patient Consents

The Medical Ethics Committee of the Academic Medical Center in Amsterdam waived the need for ethical approval. Our RCT was registered at the Netherlands Trial Register, number NTR1270. Informed consent was obtained from all participating patients and all caregivers.

Clusters

We invited all 43 Dutch multidisciplinary ALS teams (clusters) to participate. Pairs of teams were formed that were matched on three factors that may affect the QOC: the number of patients with ALS the team supports during one year (≤10; 11–20; >20); the type of center the team is working in (outpatient department of a hospital or rehabilitation center); and whether or not the team is certified by the Dutch Association for Patients with Neuromuscular Disease as a “Neuromuscular Rehabilitation Center” 11. Finally, a researcher (HC) performed computer generated randomization of the teams. Allocation concealment was at the cluster level.

Participants

The rehabilitation medicine consultant of each participating ALS team introduced the study to the eligible patients and their most important informal caregiver. Patients were not eligible for trial entry if they met one or more of the exclusion criteria: cognitive dysfunction (Mini Mental State Examination <20, 12 insufficient mastery of the Dutch language, and institutionalization. The exclusion criterion for caregivers was insufficient mastery of the Dutch language.

Blinding

In the intervention and control groups, the case manager administered the outcome assessment questionnaires during baseline home visits; assessments were unblinded for group assignment. Two independent researchers who were blinded to group assignments performed outcome assessments during follow-up at 4, 8, and 12 months.

Interventions

Usual care

Usual care in the Netherlands is neuropalliative care by multidisciplinary, secondary care teams. Such teams consist of a rehabilitation medicine consultant, an occupational therapist, physical therapist, speech pathologist, dietician, social worker, psychologist and consultant physicians (in neurology, respiratory and gastroenterology). Community and social services also play an important role in care for patients with ALS and their caregivers. General practitioners, district nurses, home care services, paramedics, social workers, and voluntary workers participate in these services.
Case management

We performed the intervention “case management” at the individual participant level. The predominant focus of the patient advocacy case management model is more comprehensive coordination of services across the continuum of care, viewed from the patient perspective.

During the 12-month intervention period, case management was provided by 2 experienced occupational therapists, specialized in ALS care and trained in client-centered practice, who used a client-centered approach to guide the participants. The case manager had an independent position outside, but in close contact with, the ALS team. The case manager’s role was to be attentive to the needs of the participants. The case manager provided participants all of the information needed to allow individual choices about how their needs would be met.

The case manager started the intervention by visiting participants at home at study entry and subsequently every three months. Between visits, contact was possible by telephone, e-mail, or in writing. At the first visit, the case manager provided participants with additional oral and written information about the procedures and objectives of the case management intervention. The position, tasks, and responsibilities of the case manager in relation to the participants’ usual care system were also elucidated.

The starting point for the case management intervention was any somatic, psychosocial, environmental, or care issue raised by the participants. On the basis of the issue, the case manager discussed the aims to achieve and the steps necessary for achievement (client-centered approach). In response to the issues raised, the case manager undertook one or more of the following steps:

- Inform participants’ multidisciplinary ALS team and other professional caregivers involved.
- Provide participants with written and oral information and advice.
- Give emotional support to participants.
- Refer participants to health care providers and agencies.
- Mediate if problems arose between participants and health care providers or agencies.
- Support participants during their contact with agencies and suppliers.
- Evaluate the handling of the issues raised by participants.

During the quarterly home visits, the case manager monitored the participants’ disease process with the aim to identify symptoms or problems related to ALS progression or caregiver burden. For that purpose, a symptom checklist was used to monitor disease progression and to register changes in disease symptoms. When the case manager noticed QOC problems or observed an increase in symptom burden, psychosocial problems, or caregiver burden, the issue was discussed with the participants. Subsequently, steps toward solutions of the relevant issues were discussed and put into action, if desired by the participants.
After randomization, the case manager and the rehabilitation medicine consultant (as coordinator of the team) agreed upon lines of communication with the ALS team during the study. The case manager visited the ALS team before initiation of the study to meet the team upon request. The case manager reported any questions of the participants, and actions or issues noticed by the case manager to the rehabilitation medicine consultant.

**Outcome measures**
The primary outcome was each patient’s health-related QOL assessed with the 40-item ALS Assessment Questionnaire (ALSAQ-40), Emotional Functioning domain (EF). The ALSAQ-40 contains 40 items incorporated in five distinct areas of health: Physical Mobility, Activities of Daily Living/Independence, Eating and Drinking, Communication, and Emotional Functioning. Each of the five scales is transformed into a scale of 0 to 100 (100 indicating worst health) \(^{18-21}\).

Secondary outcomes were caregiver strain assessed with the Caregiver Strain Index (CSI) \(^{22}\) and Quality of Care (QOC). To score QOC, we asked patients rate the total care provided on a 10-point scale, with 1 as very poor QOC and 10 as excellent QOC. Caregivers were asked to give two ratings, one for the care provided to the patient, and one for the care provided to the caregiver.

We monitored disease progression with the ALS Functional Rating Scale-Revised (ALSFRS-R). The ALSFRS-R is a 48-point validated disease-specific measure to assess function \(^{23}\); a lower score indicates more disability.

**Data analysis**
A sample size of 65 patients with ALS and their caregiver in each treatment group was planned. This group size has 80% power to detect a treatment difference of 11.9 points (SD: 26.25) in QOL scores on the ALSAQ-40 EF between the groups \(^{20}\). This calculation is based on 4 repeated measurements with a within-person correlation coefficient (time interval 4 months) of 0.75 at a significance level of 0.05 (one-sided) \(^{24}\). We calculated the sample size without taking into account the intracluster correlation coefficient, as the number of participants per multidisciplinary ALS team is relatively small.

We compared sociodemographic characteristics of the patients and their caregivers, as well as the clinical characteristics of the patients in both groups (participating vs non-participating and intervention vs control) with 2-tailed independent \(t\) tests (for continuous variables) and chi-squared tests (for dichotomous and categorical variables).

We used linear mixed models with an unstructured covariance type (multilevel analysis) to analyse differences in outcomes between the intervention and control group over time and to adjust for the clustering of the data \(^{25}\). In case of significant differences in baseline characteristics of patients and caregivers between the intervention and control group, we adjusted for these variables.
A linear mixed model does not require complete follow-up data from all participants. We expected incomplete follow-up owing to decease or withdrawal. Statistical analysis was carried out on an intention to treat basis and data of all included patients and caregivers, including those with incomplete sets of data were analysed. We calculated regression coefficients for the effects of case management compared with usual care as β values and standard errors (SE). Because outcomes are measured at the individual patient level, the analysis must be adjusted for potential clustering in the data. The analysis yields maximum likelihood estimates for the effects of time, treatment group, and the time-treatment group interaction. All hypotheses were tested 2-sided, with a critical value of <0.05.

RESULTS

Cluster randomization
Thirty-one multidisciplinary ALS teams (72%) agreed to participate in the study (figure 1). We evaluated 16 intervention teams and 15 control teams. No teams dropped out during the intervention period. Participating teams were representative of all Dutch ALS teams, and their service areas cover all parts of the country. The median cluster size was 4 patients for the intervention teams (range 0−10 patients) and 2 patients for the control team (range 0−17 patients).

Participants
From March 2009 to July 2011, 132 patients and 126 of their caregivers participated: 71 in the intervention group and 61 in the control group (figure 1). Baseline sociodemographic and clinical characteristics of patients and caregivers did not differ between the case management and control group (table 1) or between participants and non-participants (supplementary table e-1). Most caregivers were partners (n=112).

During follow-up, the proportion of missing assessments did not differ significantly between the intervention group (patients 111/284, caregivers 100/264) and the control group (patients 92/244, caregivers 98/240). Reasons for missing outcome data were patient death, patient withdrawal, and patient request to skip an assessment. The required number of patients according to the study protocol (n=130) was reached.
Figure 1 Flow of clusters and individual participants.

Enrollment of ALS teams
ALS teams invited to participate the study n=44
No consent n=13

Allocation of ALS teams
Randomization of ALS teams n=31

Intervention teams n=16
Patients n=155

Control teams n=15
Patients n=143

ALS patients No consent n=166

Enrollment:
Patients n=71
Caregivers n=66

Baseline

Enrollment:
Patients n=61
Caregivers n=60

Analysis

Completed the trial:
Patients n=43
Caregivers n=41
Death: n=23
Patient’s death: n=22
Stop: n=5
Stop: n=3

Number of evaluations of:
Patients n=173
Caregivers n=164

Completed the trial:
Patients n=36
Caregivers n=35
Death: n=15
Patient’s death: n=15
Stop: n=10
Stop: n=10

Number of evaluations of:
Patients n=152
Caregivers n=142

ALS = amyotrophic lateral sclerosis.
### Table 1 Baseline characteristics of study participants.

<table>
<thead>
<tr>
<th>Patient variables</th>
<th>Intervention (n=71)</th>
<th>Control (n=61)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age (SD), y</td>
<td>63 (11)</td>
<td>62 (11)</td>
<td>0.680</td>
</tr>
<tr>
<td>Male</td>
<td>57</td>
<td>64</td>
<td>0.786</td>
</tr>
<tr>
<td>Married</td>
<td>83</td>
<td>84</td>
<td>0.789</td>
</tr>
<tr>
<td>Primary caregiver is partner</td>
<td>82</td>
<td>84</td>
<td>0.474</td>
</tr>
<tr>
<td>Caucasian ethnicity</td>
<td>100</td>
<td>97</td>
<td>0.307</td>
</tr>
<tr>
<td>College or university degree</td>
<td>42</td>
<td>31</td>
<td>0.190</td>
</tr>
<tr>
<td>Employed</td>
<td>7</td>
<td>15</td>
<td>0.168</td>
</tr>
<tr>
<td>Limb onset</td>
<td>73</td>
<td>79</td>
<td>0.466</td>
</tr>
<tr>
<td>Median time since onset (Q1-Q3), y</td>
<td>1.8 (1.2 – 3.5)</td>
<td>2.2 (1.2 – 3.9)</td>
<td>0.600</td>
</tr>
<tr>
<td>Median time since diagnosis (Q1-Q3), y</td>
<td>0.9 (0.3 – 1.7)</td>
<td>0.9 (0.4 – 2.3)</td>
<td>0.277</td>
</tr>
<tr>
<td>Mean predicted FVC % (SD)</td>
<td>86 (23)</td>
<td>83 (24)</td>
<td>0.627</td>
</tr>
<tr>
<td>PEG</td>
<td>14</td>
<td>21</td>
<td>0.275</td>
</tr>
<tr>
<td>NIV/IV</td>
<td>17</td>
<td>18</td>
<td>0.864</td>
</tr>
<tr>
<td>Riluzole</td>
<td>91</td>
<td>95</td>
<td>0.398</td>
</tr>
<tr>
<td>Mean ALSFRS-R (SD)</td>
<td>32 (8)</td>
<td>32 (9)</td>
<td>0.685</td>
</tr>
<tr>
<td>Mean ALSAQ-40 Emotional Functioning (SD)</td>
<td>21 (18)</td>
<td>19 (17)</td>
<td>0.524</td>
</tr>
<tr>
<td>Mean ALSAQ-40 sum score (SD)</td>
<td>37 (17)</td>
<td>37 (18)</td>
<td>0.919</td>
</tr>
<tr>
<td>Mean quality of care rating score (SD)</td>
<td>8.2 (0.8)</td>
<td>8.3 (0.8)</td>
<td>0.803</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Caregiver variables</th>
<th>Intervention (n=66)</th>
<th>Control (n=60)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age (SD), y</td>
<td>59 (14)</td>
<td>57 (15)</td>
<td>0.922</td>
</tr>
<tr>
<td>Female</td>
<td>70</td>
<td>62</td>
<td>0.342</td>
</tr>
<tr>
<td>Married</td>
<td>94</td>
<td>90</td>
<td>0.463</td>
</tr>
<tr>
<td>Caucasian ethnicity</td>
<td>97</td>
<td>95</td>
<td>0.463</td>
</tr>
<tr>
<td>College or university degree</td>
<td>37</td>
<td>27</td>
<td>0.197</td>
</tr>
<tr>
<td>Employed</td>
<td>23</td>
<td>32</td>
<td>0.305</td>
</tr>
<tr>
<td>Mean CSI (SD)</td>
<td>6 (3)</td>
<td>5 (3)</td>
<td>0.184</td>
</tr>
<tr>
<td>Mean quality of care score “care for the patient” (SD)</td>
<td>8.0 (0.9)</td>
<td>8.2 (1.1)</td>
<td>0.298</td>
</tr>
<tr>
<td>Mean quality of care score “care for the caregiver” (SD)</td>
<td>7.6 (1.4)</td>
<td>7.8 (1.6)</td>
<td>0.436</td>
</tr>
</tbody>
</table>

Abbreviations: ALSAQ-40 = 40-item Amyotrophic Lateral Sclerosis Assessment Questionnaire; ALSFRS-R = Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised; CSI = Caregiver Strain Index; FVC% = percentage of the predicted value of forced vital capacity; IV = invasive ventilation; NIV = noninvasive ventilation; PEG = percutaneous endoscopic gastrostomy; Q1-Q3 = first quartile to third quartile; SD = standard deviation; y = years.

Values are percentages unless otherwise indicated.

### Case management

The duration of home visits ranged from 60 to 180 minutes. The extent to which patients and their caregivers relied on case management varied widely. Participants differed in their need for information, support, treatment, resources, and care (Supplementary Figure e-1). Seventeen patients (24%) and their caregivers did not raise any questions for the case
manager. Twenty-nine participants contacted the case manager at least once during the interval between visits. Thirty-one caregivers and 48 patients raised at least one issue during the intervention period.

Actions by the case manager were mostly related to emotional well-being (coping with loss, emotions, dependence and changes, caregiver strain, and private life) and consisted of providing a listening ear for participants, discussing the impact of having ALS and the impact of symptoms on daily functioning. Practical actions included providing oral and written information, reference to health care providers, and contacting/informing health care providers.

_Effectiveness of case management_

Despite significant disease progression, emotional well-being as reflected by ALSAQ-40 EF did not change over time. No significant difference in ALSAQ-40 EF score was detected between the intervention group and the usual care group (table 2 and figure 2).

Caregiver strain (CSI) increased significantly during the study period in both groups, without differences between the groups.

At baseline, patients as well as caregivers rated the QOC as high (table 2). The QOC scores did not change over time, and the intervention did not significantly affect the QOC rating score.
<table>
<thead>
<tr>
<th>Outcome measures</th>
<th>Case managementa Mean (SD), n</th>
<th>Usual carea Mean (SD), n</th>
<th>Linear Mixed Model β (SE); p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Average</td>
<td>Total</td>
<td></td>
</tr>
<tr>
<td>1. ALSAQ-40 Emotional Functioning (0 – 100)b</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>21.3 (18.2), 70</td>
<td>19.3 (17.0), 61</td>
<td>Time: 0.54 (0.58) p = 0.35</td>
</tr>
<tr>
<td>4 months</td>
<td>19.8 (14.6), 43</td>
<td>19.4 (16.6), 39</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>21.5 (13.5), 28</td>
<td>20.0 (16.1), 22</td>
<td>Time x Group: 0.33 (0.79) p = 0.68</td>
</tr>
<tr>
<td>12 months</td>
<td>22.8 (16.4), 30</td>
<td>19.1 (14.7), 27</td>
<td></td>
</tr>
<tr>
<td>2. CSI (0-13)b</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>5.8 (3.4), 66</td>
<td>5.0 (2.9), 59</td>
<td>Time: 0.60 (0.11) p &lt; 0.0001c</td>
</tr>
<tr>
<td>4 months</td>
<td>7.1 (3.4), 42</td>
<td>6.4 (3.1), 37</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>7.0 (3.6), 25</td>
<td>7.4 (3.5), 20</td>
<td>Time x Group: -0.08 (0.15) p = 0.59</td>
</tr>
<tr>
<td>12 months</td>
<td>7.9 (2.9), 29</td>
<td>7.3 (3.2), 24</td>
<td></td>
</tr>
<tr>
<td>3. QOC report mark patients with ALS (1 – 10)d</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>8.2 (0.8), 65</td>
<td>8.3 (0.8), 55</td>
<td>Time: -0.06 (0.04) p = 0.13</td>
</tr>
<tr>
<td>4 months</td>
<td>8.2 (0.9), 43</td>
<td>8.0 (0.7), 37</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>8.1 (1.0), 28</td>
<td>8.0 (1.1), 23</td>
<td>Time x Group: -0.02 (0.05) p = 0.69</td>
</tr>
<tr>
<td>12 months</td>
<td>7.9 (0.9), 30</td>
<td>8.0 (0.7), 25</td>
<td></td>
</tr>
<tr>
<td>4. QOC report mark caregivers: patient care (1 – 10)d</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>8.0 (0.9), 63</td>
<td>8.2 (1.1), 59</td>
<td>Time: -0.05 (0.04) p = 0.21</td>
</tr>
<tr>
<td>4 months</td>
<td>8.3 (0.8), 40</td>
<td>8.2 (0.8), 37</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>8.0 (0.9), 26</td>
<td>8.1 (0.8), 18</td>
<td>Time x Group: 0.02 (0.06) p = 0.21</td>
</tr>
<tr>
<td>12 months</td>
<td>7.9 (0.7), 27</td>
<td>7.9 (0.9), 24</td>
<td></td>
</tr>
<tr>
<td>5. QOC report mark caregivers: caregiver care (1 – 10)d</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>7.6 (1.4), 56</td>
<td>7.8 (1.6), 49</td>
<td>Time: -0.10 (0.06) p = 0.12</td>
</tr>
<tr>
<td>4 months</td>
<td>7.7 (1.0), 38</td>
<td>7.7 (1.2), 33</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>7.3 (1.6), 25</td>
<td>7.6 (1.0), 16</td>
<td>Time x Group: 0.004 (0.08) p = 0.96</td>
</tr>
<tr>
<td>12 months</td>
<td>7.3 (1.3), 25</td>
<td>7.1 (1.0), 22</td>
<td></td>
</tr>
<tr>
<td>Disease progression: ALSFRS-R (0 – 48)d</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>32.1 (7.9), 71</td>
<td>31.6 (8.8), 61</td>
<td>Time: -2.64 (0.40) p &lt; 0.0001c</td>
</tr>
<tr>
<td>4 months</td>
<td>29.9 (7.0), 43</td>
<td>27.6 (10.2), 37</td>
<td></td>
</tr>
<tr>
<td>8 months</td>
<td>27.4 (7.1), 27</td>
<td>27.8 (11.0), 20</td>
<td>Time x Group: -0.26 (0.54) p = 0.63</td>
</tr>
<tr>
<td>12 months</td>
<td>24.0 (9.3), 28</td>
<td>25.1 (11.5), 25</td>
<td></td>
</tr>
</tbody>
</table>

Abbreviations: ALSFRS-R = Amyotrophic Lateral Sclerosis Functional Rating Scale Revised; ALSAQ-40 = ALS Assessment Questionnaire-40 items; CSI = Caregiver Strain Index; QOC = Quality Of Care; SD = standard deviation; SE = standard error.

β = multilevel regression coefficient for the effect of case management compared with usual care, and is an estimation of the longitudinal relationship between the parameter and the outcome.

Time = change over time (4 months) in outcome for both groups; Time x treatment group interaction term = change over time (4 months) in outcome different for both groups.

a Intracluster correlation coefficient (ICC): All ICC values were less than zero.

b Score range; lower scores indicate a better situation.

c p < 0.05.

d Score range; higher scores indicate a better situation.
DISCUSSION

This cluster randomized clinical trial on case management in multidisciplinary ALS care demonstrated no significant benefit of case management to the emotional functioning of patients, the level of strain experienced by the caregiver, or the level of satisfaction of patients and caregivers when added to standard multidisciplinary care compared with
standard care alone. The design and results of our study must be viewed in the context of the Dutch ALS care setting. Participants were recruited from ALS teams, and all received standard multidisciplinary ALS care. Previous studies demonstrated that multidisciplinary ALS care results in a better QOL and satisfaction with QOC than non-specialized general care. We estimate that at least 80% of patients with ALS are presently being referred to a multidisciplinary ALS team in the Netherlands. To further explore the effect of case management models in ALS, future studies must be conducted in other health care systems.

In other diseases, effectiveness of case management on patients’ QOL has been demonstrated, although the evidence is limited due to inconsistent findings.

The present study aimed to improve any shortcomings in the needs of patients with ALS and their caregivers. Judging from the frequency of questions and issues raised during the intervention period, the intervention did not result in an intensive use of case management by the participants. The high scores on mental QOL and satisfaction with care and the modest number of issues raised by patients and their caregivers suggest that problems in Dutch multidisciplinary ALS care, as identified in daily practice, are incidental, with relatively low impact on mental QOL and overall satisfaction with ALS care.

In our study design, we deliberately chose a case manager who was not a member of the multidisciplinary ALS team. Patients and caregivers in this way had the opportunity to consult the independent case manager for a second opinion or when the ALS team did not adequately address the needs of patients/caregivers. Regular contacts between the case manager and the team were assured to avoid overlap. The case managers’ professional background in our opinion did not influence our study results: their broad overview of the Dutch ALS care and their extensive experience in ALS care were appropriate for their signaling, supportive and advisory function.

We chose an ALS-specific health-related QOL measure to evaluate the effect of case management. SMiLE and SEIQOL measure non-health-related factors of QOL and are more focused on psychological, existential and support factors. We cannot exclude an effect on those measures. However, the lack of effect does not seem to be due to the scales used as we did not find an effect on any other outcome measure.

In addition to the lack of effectiveness for patients with ALS, we were unable to demonstrate any improvement in caregivers’ burden. The increased caregiver burden over time has been described earlier. This finding may result from the fact that most issues raised during the intervention period were directly aimed at patients and were less often issues concerning caregiver strain. This possibility is illustrated by the content of the issues raised, which was mostly related to the provision of assistive products and technology, restrictions in activities and participation, symptom treatment, psychosocial problems, and provision of palliative care, illustrating that the focus was more on the patient than the caregiver. Our finding of an increasing burden for caregivers indicates the need for a more prominent role of interventions targeting caregivers in future studies.
The strengths of this study are its randomized controlled design and the large sample size of patients with ALS, which is representative for the Dutch ALS population as we compare patient characteristics (with respect to age at disease onset, sex, time to diagnosis, and site of onset) with those of a recently described population-based epidemiology study. In addition, we applied multilevel analysis to account for various potential confounding factors.

The number of missing data could be considered a limitation of the study, but is inherent to longitudinal studies of patients with ALS. Loss to follow-up is not expected to have a substantial effect on the power of the study considering the size of the multilevel regression interaction coefficient for the effect of case management. Irrespective of the number of observations per participant, every participant was included in the multilevel analyses, and these analyses deal appropriately with the varying numbers of observations. Another possible limitation of this study is the limited duration of the intervention period (12 months). Wynia et al. (2010) stated that although there is no evidence, it seems reasonable to assume that an “investment effect” is applicable for outcome variables such as QOL. However, 12 months is relatively long considering that ALS care is intensive during a relatively short disease period compared with other progressive diseases (e.g., multiple sclerosis). It is credible that the quarterly home visits, combined with the possibility of contact by telephone and e-mail between visits, allowed for our intervention period to be long enough to build trust and stability in the case manager/participant relationship.

Our data do not support the implementation of case management as part of ALS care. Because health care systems vary considerably between countries and regions, as does the rate of implementation of multidisciplinary ALS care, our research design facilitates future case management research projects in other countries, as well as outside the context of specialized ALS teams. Such studies will decisively determine the effect of different case management models in ALS care.

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REFERENCES


17. Client-centred practice: what does it mean and does it make a difference?


Supplementary Table e-1 Baseline characteristics of study participants versus non-participants.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Participants (n=132)</th>
<th>Non-participants (n=166)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age (SD), years</td>
<td>62.9 (11.2)</td>
<td>63.4 (11.7)</td>
<td>0.702</td>
</tr>
<tr>
<td>Male</td>
<td>65</td>
<td>57</td>
<td>0.151</td>
</tr>
<tr>
<td>Married</td>
<td>83</td>
<td>77</td>
<td>0.375</td>
</tr>
<tr>
<td>Primary caregiver is partner</td>
<td>83</td>
<td>67</td>
<td>0.312</td>
</tr>
<tr>
<td>Caucasian ethnicity</td>
<td>99</td>
<td>96</td>
<td>0.326</td>
</tr>
<tr>
<td>College or university degree</td>
<td>42</td>
<td>30</td>
<td>0.190</td>
</tr>
<tr>
<td>Limb onset</td>
<td>76</td>
<td>70</td>
<td>0.233</td>
</tr>
<tr>
<td>Median time since onset (Q1-Q3), years</td>
<td>2.1 (0.3 - 12.6)</td>
<td>2.3 (0.1 - 22.7)</td>
<td>0.372</td>
</tr>
<tr>
<td>Median time since diagnosis (Q1-Q3), years</td>
<td>0.9 (0.05 - 12.1)</td>
<td>1.1 (0.01 - 22.2)</td>
<td>0.446</td>
</tr>
<tr>
<td>Mean predicted FVC % (SD)</td>
<td>85 (22)</td>
<td>81 (21)</td>
<td>0.325</td>
</tr>
<tr>
<td>PEG use</td>
<td>17</td>
<td>19</td>
<td>0.680</td>
</tr>
<tr>
<td>NIV/IV use</td>
<td>17</td>
<td>13</td>
<td>0.273</td>
</tr>
<tr>
<td>Riluzole</td>
<td>93</td>
<td>90</td>
<td>0.326</td>
</tr>
<tr>
<td>Median ALSFRS-R (Q1-Q3)</td>
<td>33 (26 – 39)</td>
<td>32 (26 – 40)</td>
<td>0.868</td>
</tr>
</tbody>
</table>

Abbreviations: ALSFRS-R = Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised; FVC% = percentage of the predicted value of forced vital capacity; IV = invasive ventilation; NIV = noninvasive ventilation; PEG = percutaneous endoscopic gastrostomy; Q1-Q3 = first quartile to third quartile; SD = standard deviation.

Values are percentages unless otherwise indicated.

Supplementary Figure e-1 Themes of issues about which participants raised questions.

Number of ALS patients and/or caregivers who raised the issue

Provision of assistive products and technology: 39
Activities and participation: 38
Symptoms: 37
Psychosocial: 35
Palliative care provision: 31
Personal care services: 19
Finances: 19
Nutrition / PEG: 13
Domestic life services: 12
Transportation services: 10
Advance directives / end of life decisions: 10
Respiratory care / NIV: 9
Work: 7
Durable power of attorney: 7
Respite care: 5
Supporting children: 3
Social services / voluntary work: 2

Abbreviations: PEG, percutaneous endoscopic gastrostomy; NIV, non-invasive ventilation.