Abstract

**Background:** Physical therapy is an essential component of multidisciplinary treatment in amyotrophic lateral sclerosis (ALS). However, the meaning of physical therapy beside preservation of muscular strength and functional maintenance is not fully understood.

**Objective:** The purpose of this study was to examine the patient’s perception of physical therapy during symptom progression using an internet assessment approach.

**Methods:** A prospective, longitudinal, observational study was performed. Recruitment took place in an ALS center in Berlin, Germany. Online self-assessment was established on a case management platform over 6 months. Participants self-assessed the disease progression using the ALS Functional Rating Scale, revised (ALSFRSr). The specific target of physical therapy was rated by Measure Yourself Medical Outcome Profile (MYMOP). To inquire about the level of recommendation we used the net promoter score (NPS).
**Results:** 45 participants with ALS were included. 27 participants (60 %) started the online assessment. The mean duration of physical therapy sessions per week was 143 minutes (SD 60.4) with a mean frequency of 2.9 (SD 1.2) per week. As defined by MYMOP, most concerning symptoms were reported in the lower extremities (62 %), in the upper limbs (31 %) and less frequently in the trunk (7 %). Notwithstanding physical therapy, there was a functional decline of 3 points in the ALSFRSr at the end of the observation (n = 20). Furthermore, the MYMOP showed a significant loss of 0.8 points in the composite score, 0.9 points in the activity score and 0.8 points in the targeted symptom. Contrary to the functional decline, recommendation of PT raised from a baseline value of 20 NPS points to very high 50 points at the end of study ($P = 0.05$).

**Conclusion:** Physical therapy is perceived as an important treatment by patients with ALS. Despite a functional decline, patients are satisfied with their physical therapy and recommend this intervention. The results underline the changing meaning of physical therapy throughout the course of the disease. Physical therapy in ALS has to be regarded as a supportive and palliative health care intervention beyond functional outcome parameters.

**Keywords:** amyotrophic lateral sclerosis, physical therapy, MYMOP, net promoter score (NPS), online self-assessment
Introduction

Amyotrophic lateral sclerosis (ALS) is a fatal neurodegenerative disease characterised by a loss of motor neurons in the cortex, brainstem and spinal cord which results in a progressive motor deficits and a paralysis of the muscles controlling the limbs, swallowing and breathing abilities [1]. With disease progression muscles of fine and gross motor functions are affected, leading to a decline of motor skills and activities. As there is no current curative treatment for ALS, the focus of the management of the complex symptoms lies on multidisciplinary care. It includes symptomatic, rehabilitative and palliative therapy, delivered by an interprofessional team that consists of different fields such as neurologists, nurses, and therapists, working in a coordinated and organised manner [2]. An important part of multidisciplinary treatment is physical therapy, which is widely prescribed and applied in the treatment of ALS. In a European survey, it has been shown that physical therapy is performed in 83 % of ALS patients [3]. Physiotherapists play an essential role in the multidisciplinary care team emphasizing on improving function and quality of life in patients who are deemed to require physical and functional dimensions of palliative care [4].

Experimental data [5-7] and several randomised trials showed moderate effects and benefits of submaximal resistive exercises especially in early stages of the disease [8,9]. The neuromuscular mechanism was thought to be prevention of disuse atrophy and more efficient motor unit recruitment. Excessive or high resistant exercises have been associated with overwork damage and thus are not recommended in ALS [8]. The key focus for physical therapists is to delay the decline of muscular strength by submaximal resistance exercise, which has been shown to be safe and efficacious.

However, there is still an uncertainty about the manner, duration and frequency of physical therapy. This immanent lack of defined treatment guidelines apparently arises from the encounter of a large clinical heterogeneity in ALS syndromes, different therapeutic approaches and individual expectations of patients and therapists.

Thus, the aims of the present study were to (I) evaluate the frequency and duration of physical therapy sessions among ALS patients, (II) to determine the symptoms most bothering, and (III) to identify the recommendation of physical therapy and the Net Promoter Score (NPS) at the beginning and end of the study. We hypothesized that the recommendation of physical therapy decreases with symptom progression in ALS.
Furthermore, we hypothesized that recommendation of physical therapy is related to the most bothering symptom, disease severity, duration, or frequency of physical therapy sessions in ALS.

**Methods**

**Study Design and Recruitment**

This is a prospective, longitudinal, observational study that recruited a consecutive cohort of participants from the ALS outpatient department at Charité – Universitätsmedizin Berlin, Germany. Baseline assessment of epidemiological data, symptoms, type and amount of physical therapy has been performed with 45 individuals, whereas 20 participants completed an online survey over a period of 6 months according symptom severity, restriction of activity and recommendation of physical therapy.

**Setting**

For online self-assessment and evaluation of physical therapy the digital and internet-supported case management network Ambulanzpartner Soziotechnologie (APST) was used [10]. The internet platform of APST encompassed the service of coordinators specialized in case management, a tailored digital management platform and assessment tools for self-assessment, services, therapy and assistive devices [11]. The patients and their caregivers were granted access to the APST platform through an account.

**Participants**

Inclusion criteria for this study were a possible, probable or definitive diagnosis of ALS following the revised El Escorial criteria [12], a stage of a disease were at least one motor function was restricted and a participation in physical therapy. Patients with other severe life limiting diseases or who showed clinical significant cognitive impairment were not eligible for this trial. For online assessment participants entered the digital case management program provided by APST [10].
Variables and data sources

Physical Therapy

Physical therapy was prescribed by a neurologist specialized in ALS and realized by physical therapists trained in the treatment of neurological disorders including ALS. In addition to the prescription of physical therapy patients received special treatments like massages, lymphatic drainage, thermal, or breathing therapy if needed. The overall time per week and frequency of physical therapy sessions alone and in addition with special treatments was documented.

ALSFRSr

We evaluated the functional impairment of the participants using the ALS functional rating scale, revised (ALSFRSr) as an online self-assessment [13]. This score is a validated and widely used instrument that comprises the fine and gross motor functions of the upper and lower extremities, bulbar functions and breathing abilities. It comprises 12 items of short, clear questions with well-defended five anchor points (0-4) for response options. Hence, the range of the total scale spans from 0 to 48 score points, where less points represents more severe symptoms. The loss of ALSFRSr value per month, or delta ALSFRSr, indicates the rate of deterioration and predicts survival [14].

MYMOP

To focus on a single specific most bothersome or disabling motor symptom we employed the Measure Yourself Medical Outcome Profile (MYMOP) [15, 16]. The MYMOP is a brief, patient generated, problem specific questionnaire, which requires the participants to specify the symptom which is concerning them most. Subsequently, participants evaluate the severity of this symptom on a 7-point Likert scale (e.g. weakness of the right leg from 0 = ‘as good as it could be’ to 6 = ‘as bad as it could be’) related to the previous week. In a second part, the limitation of an activity of daily living or a movement that is being restricted or prevented by the symptom (e.g. walking) is assessed on the same scale. They also score their well-being. Follow-up questionnaires address the original concerns. All domains (symptom severity, restriction of activity, and well-being) can be analysed individually or as a total score, the profile score, that equals the mean of the sub scores recorded (score 0-6).
To evaluate the overall recommendation of physical therapy, we used a numeric rating scale (NRS) that derives from the net promoter score (NPS) [17, 18], which is used in customer relation management and has been introduced to clinical assessment recently [19, 20]. The NPS is an easy-to-use, one-item questionnaire that is based on the question “How likely is it, that you would recommend the service to a friend or colleague?” Participants were asked to score on a 0 to 10 NRS, where 10 means extremely likely to recommend the therapy. To calculate the NPS, the percentage of participants who respond 0 – 6 is subtracted from the percentage of those who scored 9 – 10. Participants with the values 7 and 8 are assumed to be indifferent or passive. Therefore, the NPS can be as low as −100 were everybody would be a detractor or as high as +100 were everybody would be a promoter. A positive NPS is supposed to be good, whereby an NPS of more than 50 % is considered excellent. Alternatively, to avoid the problem of the NPS categorization, it could be considered to refrain from calculating the Net Promoter Score and only report the average NPS [18].

Data Analysis
Data were analysed with IBM SPSS Statistics (Version 24.0) for Windows. Results were expressed as means (± SD) if normally distributed and medians (maximum/minimum) if distribution was non-Gaussian. Correlational analysis was performed with Spearman’s rho because of the ordinal nature of the scales. A statistically significant difference of paired samples was analysed with a t-test. Recommendation was tested with the Wilcoxon test for related samples. A P value of < .05 (two-tailed) was considered significant.

Protocol approvals and registrations
The study protocol was approved by the Medical Ethics Committee of the Charité – Universitätsmedizin Berlin, Germany. A data safety and monitoring board supervised the study. A signed patient information and informed consent form was obtained from all participating patients.

Results
Descriptive data
45 participants have been included in this study and performed the baseline assessment. 27 also consented to the online assessment of MYMOP and recommendation of physical
therapy over a period of 20 weeks. 20 participants finished the 20-week online assessment providing complete data sets (Figure 1).

![Flowchart of participants](image)

Figure 1: Flowchart of participants

The mean age of all participants at baseline was 59 years (SD = 10.6) with a relatively long disease duration of 27 months (median; min/max: 3/203) due to a higher percentage of long term survivors in our trial. The mean duration of PT was 142.7 minutes (SD = 60.4) per week and mean frequency was 2.9 sessions per week (SD = 1.2). Occupational therapy and speech and language therapy (SLP) are not included in these values.

The demographics and baseline characteristics of the participants are presented in Table 1.

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Cohort 1 (all patients) at baseline</th>
<th>Cohort 2 (onsite plus online) at baseline</th>
<th>Cohort 2 (onsite plus online) dropouts</th>
<th>Cohort 2 (onsite plus online) at study end</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(n = 45)</td>
<td>(n = 27)</td>
<td>(n = 7)</td>
<td>(n = 20)</td>
</tr>
<tr>
<td>Age (yrs), mean (SD)</td>
<td>59.2 (10.6)</td>
<td>59.4 (11.1)</td>
<td>58.0 (5.6)</td>
<td>59.9 (12.5)</td>
</tr>
<tr>
<td>Gender, female/male (%)</td>
<td>16/29 (36/64)</td>
<td>7/20 (35/65)</td>
<td>3/4 (43/57)</td>
<td>4/16 (20/80)</td>
</tr>
<tr>
<td>ALSFRSr(^a) baseline, mean (SD)</td>
<td>36.9 (6.9)</td>
<td>38.5 (4.8)</td>
<td>36.1 (5.0)</td>
<td>39.4 (1.0)</td>
</tr>
<tr>
<td>delta ALSFRSr(^b), mean (SD)</td>
<td>.57 (.51)</td>
<td>.61 (.57)</td>
<td>.86 (.27)</td>
<td>.53 (.11)</td>
</tr>
<tr>
<td>Duration of disease in months</td>
<td>27.0 (3/203)</td>
<td>25.0 (3/194)</td>
<td>16.0 (9/87)</td>
<td>26.5 (3/195)</td>
</tr>
<tr>
<td>median (min/max)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total MYMOP(^c) baseline, mean (SD)</td>
<td>3.0 (SD 0.9)</td>
<td>3.0 (SD 0.9)</td>
<td>3.1 (SD 1.1)</td>
<td>2.9 (SD 0.9)</td>
</tr>
<tr>
<td>PT(^e) time in minutes per week,</td>
<td>142.7 (60.4)</td>
<td>151.1 (63.5)</td>
<td>157.1 (70.4)</td>
<td>149.0 (62.7)</td>
</tr>
</tbody>
</table>
Table 1: Baseline characteristics of participants

<table>
<thead>
<tr>
<th></th>
<th>Mean (SD) 1</th>
<th>Mean (SD) 2</th>
<th>Mean (SD) 3</th>
<th>Mean (SD) 4</th>
</tr>
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<tbody>
<tr>
<td>Overall time prescribed</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Including special treatments,</td>
<td>269.3 (138.6)</td>
<td>263.6 (110.7)</td>
<td>216.6 (58.5)</td>
<td>280.1 (120.7)</td>
</tr>
<tr>
<td>PT frequency per week, mean</td>
<td>2.9 (1.2)</td>
<td>2.9 (1.2)</td>
<td>2.9 (1.5)</td>
<td>3.0 (1.2)</td>
</tr>
</tbody>
</table>

a: standard deviation; b: ALSFRSr = Amyotrophic Lateral Sclerosis Functional Rating Scale, revised; c: loss of ALSFRSr points per month; d: MYMOP = Measure Yourself Medical Outcome Profile; e: PT = physical therapy

Main Results

Physical Therapy

There was no significant difference of the prescribed physical therapy time and session frequency in the different cohorts. Given the fact that a regular physical therapy unit lasts between 45 to 60 minutes and patients receive 3 units per week, the mean duration of therapy sessions amounts to two and a half to three hours per week. Interestingly, an additional two hours per week were granted for special treatments. Only the early terminating online participants appeared with less special treatment time of just one hour per week. However, this result did not reach statistical significance ($P = .19$).

ALSFRSr

The ALSFRSr at baseline was comparable with other trials but ALS progression rate was .57 (SD = .4) which is lower than in an average ALS population where the loss is usually .8 to 1 point. In the online cohort the constant decline in motor function is represented by an expected significant decline in the total ALSFRSr from 39.4 to 36.4 ($P = .05$). Those 18 patients who did not participate in the online assessment showed a significantly more advanced stage of the disease compared to those who attended (ALSFRSr: 34.4 vs. 38.5, $P = .05$). 7 participants withdrew from the online survey after 7.7 weeks (SD = 5.8). In these participants a higher percentage was female (57 vs. 43 %) and tended to be more
affected (ALSFRSr: 36.1 vs. 39.4; \( P = .13 \)) with a higher rate of progression (delta ALSFRSr: .72 vs .53; \( P = .19 \)).

**MYMOP**

Based on the MYMOP initial questionnaire 62% of participants defined symptoms in the lower extremities as most bothering, while restrictions in the arms were the most important issue in 31%. Axial symptoms like weakness of the trunk were stated in 7% of the participants as dominating symptom (Figure 2).

![Figure 2: Distribution of the most concerning symptom at baseline (n = 45)](image)

The total MYMOP of all cohorts at baseline was similar between 2.9 and 3.1. The profile score of MYMOP at baseline of 3.0 (SD = .9, n = 45) did not significantly correlate with the total ALSFRSr at baseline (\( r = .27; P = .17 \)). Whereas the correlation of the MYMOP with the according ALSFRSr sub score related to upper and lower extremities was significant (\( r = .45; P = .003 \)). However, the highest correlation was seen between the ALSFRSr lower extremities sub score and the MYMOP symptom assessment sub score (\( r = .62; P < .001 \)). This correlation was reproducible throughout the trial.

In the online cohort (n = 20, Table 2) the profile score of MYMOP increased from 2.9 to 3.7 (\( P = .005 \)). The MYMOP sub scores for activity increased from 3.1 to 4.0 (\( P = .02 \)). The burden of the target symptom increased from 3.1 to 3.9 (\( P = .02 \)). The wellbeing sub score displayed
a strong trend towards poorer wellbeing after 20 weeks (from 2.6 to 3.2) but without statistical significance ($P = .08$).

The seven withdrawing participants showed initially a poorer wellbeing sub score in the MYMOP compared to participant who finished the assessment (3.4 vs. 2.6; $P = .08$).

<table>
<thead>
<tr>
<th></th>
<th>Baseline</th>
<th>Week 20</th>
<th>Significance level</th>
</tr>
</thead>
<tbody>
<tr>
<td>ALSFRSr$^a$ (SD$^b$)</td>
<td>39.4 (1.0)</td>
<td>36.4 (1.3)</td>
<td>$P = .001$</td>
</tr>
<tr>
<td>MYMOP$^c$, profile (SD)</td>
<td>2.9 (.2)</td>
<td>3.7 (.2)</td>
<td>$P = .005$</td>
</tr>
<tr>
<td>MYMOP, wellbeing (SD)</td>
<td>2.6 (.3)</td>
<td>3.2 (.3)</td>
<td>$P = .080$</td>
</tr>
<tr>
<td>MYMOP, activity (SD)</td>
<td>3.1 (.2)</td>
<td>4.0 (.3)</td>
<td>$P = .020$</td>
</tr>
<tr>
<td>MYMOP, symptom (SD)</td>
<td>3.1 (.2)</td>
<td>3.9 (.3)</td>
<td>$P = .017$</td>
</tr>
<tr>
<td>Recommendation (SD)</td>
<td>7.6 (.4)</td>
<td>8.6 (.3)</td>
<td>$P = .020$</td>
</tr>
<tr>
<td>NPS$^d$ (SD)</td>
<td>20</td>
<td>55</td>
<td></td>
</tr>
</tbody>
</table>

$^a$: ALSFRSr = Amyotrophic Lateral Sclerosis Functional Rating Scale, revised; $^b$: standard deviation; $^c$: MYMOP = Measure Yourself Medical Outcome Profile; $^d$: NPS = Net Promoter Score

Table 2: Change in the online cohort over 20 weeks in the online cohort (n = 20)

**Recommendation and NPS**

The total value of recommendation of physical therapy raised from 7.6 to 8.6 ($P = .02$; Figure 3). In the 7 withdrawing participants we could see a not statistically significant decrease of recommendation based on the last assessment before withdraw (7.4 (SD = 2.2) vs. 7.0 (SD = 3.5).

The recommendation was not influenced by the following factors: age, gender, amount of physical therapy, the site of the most concerning symptom, the degree of functional impairment, wellbeing or activity (data not shown).
Figure 3: Recommendation of physical therapy at week 1 and at week 20 ($P < .05$)

Based on the recommendation we calculated the NPS, which increased from 20 at the beginning to 50 at the end of observation interval (Figure 4).

Figure 4: Change in Net Promoter Score from week 1 to week 20
Discussion

Principal findings
The aim of this observation was to determine the patients’ perception on physical therapy during disease progression. To our knowledge there are few systematic reports about the extent to which physical therapy is applied to ALS patients. Baseline assessment of this study revealed a mean duration of 270 minutes of prescribed physical therapy including special treatments and a mean frequency of three units per week. Our data did not show a significant correlation between recommending physical therapy and the extent (duration and frequency) of its application.

ALS progression measured by the ALSFRSr was complimented by the MYMOP in order to show the effect of motor decline on PT perception. The correlation of MYMOP with the motor domain of the ALSFRSr was strong, although this score is unable to measure new or several coexistent problems and concerns. The patient-centered assessment was not capable to measure all perceived benefits, like the social and psychological meanings of physical therapy.

However, the studies have found no evidence that decline in wellbeing, motor function, and level of activity significantly degrades the overall recommendation of physical therapy. Remarkably, throughout the study the rating of physical therapy improved in the majority of participants despite the functional decline. We found a good satisfaction with physical therapy at first online assessment as shown by a NPS of 20. At week 20 the NPS value reached a notable value of 50, which is considered to be excellent (20) and shows a high acceptance of physical therapy within the studied cohort.

Limitations
Our finding must be considered in the context of their limitations. Of all 45 participants, male patients were overrepresented compared to the proportion in the general ALS population. This inadequacy raised in the online cohort, which we already have observed in former online assessment trials. The approach of this study to recruit participants offline intended to reduce this bias. However, women were more likely to early terminate the assessment. The seven participants who early discontinued showed a trend towards a faster progression, a lower wellbeing and lower recommendation of physical
therapy. Presumably, a more aggressive disease progression might be a reason for dropout, as well as discontent with physical therapy. Measuring satisfaction using an online self-assessment can be challenging. The NPS allows a rating of physical therapy from a patient’s experience perspective. At the same time, it is a one dimensional item question and therefore assumed to be less reliable and more volatile than a composite index. In future studies, multidimensional or open designs should be considered to explore the patients’ perspective on physical therapy to a greater depth.

A further limitation is the single center recruitment and the small sample size. Therefore, generalization must be done with caution. Our cohort was representative to the ALS population in terms of mean age and ALSFRSr, but showed a longer mean disease duration. The progression rate of .52 (SD = .4) is lower than in an average ALS population, which is because of a wider range in the diseases courses compared to pharmaceutical trials with homogenized populations. We can imagine that patients that progress slower and long-time survivors have a certain and eventually more positive attitude towards physical therapy. Finally, our population was seen at a specialist center supported by a case management platform. Furthermore, it is located in an advanced country with a universal multi-payer health system where costs for physical therapy are covered mostly by compulsory health insurances; our findings may not generalize readily to other populations.

**Conclusion**

The overall positive rating of physical therapy cannot be fully explained with the established rehabilitative concept of physical therapy. Our data suggest an important role of physical therapy in a palliative context, where the therapy and presumably the role of the therapist itself has a considerable meaning to the patient. Physical therapists serving as interdisciplinary team members in palliative care settings, provide patient care beyond physical and bodily treatment aims. This concept includes shifting priorities across a continuum and the changing perception of physical therapist as well as other allied health specialists [21]. These palliative and multidisciplinary approaches should be encouraged during education and further training to implement the changing perceptions of PT into the qualification of physical therapists.
Acknowledgments
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Conflicts of Interest
TM received consultancy fees from Cytokinetics, GSK and Desitin Arzneimittel GmbH; served on scientific advisory boards for Cytokinetics, GSK and TEVA. TM and CM are founders of the internet platform Ambulanzpartner and hold shares of Ambulanzpartner Soziotechnologie APST GmbH.

Abbreviations
ALS: Amyotrophic Lateral Sclerosis
APST: Ambulanzpartner Soziotechnologie APST GmbH
ALSFRSr: Amyotrophic Lateral Sclerosis Functional Rating Scale, revised
MYMOP: Measure Yourself Medical Outcome Profile
NPS: Net Promoter Score
SD: standard deviation
References


