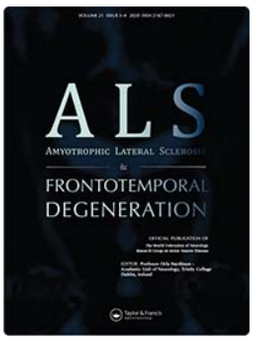

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**REVIEW ARTICLE**

# The current use of telehealth in ALS care and the barriers to and facilitators of implementation: a systematic review

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**Abstract**

*Objective*: We aimed to provide an overview of telehealth used in the care for patients with amyotrophic lateral sclerosis (ALS), and identify the barriers to and facilitators of its implementation. *Methods*: We searched Pubmed and Embase to identify relevant articles. Full-text articles with original research reporting on the use of telehealth in ALS care, were included. Data were synthesized using the Consolidation Framework for Implementation Research. Two authors independently screened articles based on the inclusion criteria. *Results*: Sixteen articles were included that investigated three types of telehealth: Videoconferencing, home-based self-monitoring and remote NIV monitoring. Telehealth was mainly used by patients with respiratory impairment and focused on monitoring respiratory function. Facilitators for telehealth implementation were a positive attitude of patients (and caregivers) toward telehealth and the provision of training and ongoing support. Healthcare professionals were more likely to have a negative attitude toward telehealth, due to the lack of personal evaluation/contact and technical issues; this was a known barrier. Other important barriers to telehealth were lack of reimbursement and cost-effectiveness analyses. Barriers and facilitators identified in this review correspond to known determinants found in other healthcare settings. *Conclusions*: Our findings show that telehealth in ALS care is well-received by patients and their caregivers. Healthcare professionals, however, show mixed experiences and perceive barriers to telehealth use. Challenges related to finance and legislation may hinder telehealth implementation in ALS care. Future research should report the barriers and facilitators of implementation and determine the cost-effectiveness of telehealth.

**Keywords:** *Amyotrophic lateral sclerosis, telehealth, barriers and facilitators, implementation*

## Introduction

Patients with amyotrophic lateral sclerosis (ALS) suffer from progressive disability, which develops at a variable rate, resulting in ever-changing care-needs. Symptomatic management by a multidisciplinary team of specialists is the mainstay of treatment for patients with ALS. This type of care aims to optimize patients' quality of life and survival (1–5). For this reason, patients should be monitored closely and have continuous access to multidisciplinary care throughout their disease. However, many patients with ALS experience issues with accessing and attending multidisciplinary clinics. These issues are mostly related to long travel distances, difficulty traveling and long days at the clinic (6,7). In addition, there is a lack of monitoring between clinic visits (in ALS care). The access issues and lack of monitoring limit the continuity of multidisciplinary care, which could negatively affect patients with ALS.

Telehealth has the potential to improve the accessibility and continuity of ALS care by enabling the remote provision of care and facilitating remote monitoring. The use of telehealth allows patients to receive specialist care, regardless of their ability to

Supplemental data for this article can be accessed here.

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Table 1. Definitions of implementation determinants.

| Domain | Construct | Definition |
|---|---|---|
| Innovation characteristics | Core components | The essential and indispensable elements of innovation. |
| | Complexity | To what extent the telehealth innovation is difficult to startup, use or implement. |
| Patient/caregiver characteristics | User-experiences and benefits | The user-experiences of the patient and caregiver with the telehealth innovation and the perceived benefits or drawbacks. |
| | Compliance | The ability or willingness of the end-user to use the telehealth innovation |
| Healthcare professional characteristics | User-experiences and benefits | The user-experiences of the healthcare professional with the telehealth innovation and the perceived benefits or drawbacks. |
| Inner and outer setting | Available resources | The available software, equipment, support, training, education and time required for operating the telehealth innovation. |
| | Finance and legislation | The available financial and legislative constructs required for operating the telehealth innovation. |

Based on the consolidation framework for implementation research.

travel, their level of impairment or the distance to a multidisciplinary clinic. Despite these potential benefits and the availability of digital technology, the use of telehealth in ALS care is currently limited. This view is supported by a recent systematic review that looked into the use of digital technology to improve access to specialist ALS care (8). The limited number of studies in the review were mostly feasibility or pilot studies and/or included only a small number of patients with ALS. This lack of (robust) literature suggests that telehealth innovations rarely survive beyond the initial pilot phase and are not implemented into usual ALS care.

These findings indicate that there are issues that hinder the implementation of telehealth in ALS care. In order to facilitate telehealth implementation, we describe its current use in ALS care and aim to identify the barriers and facilitators that influence implementation.

## Methods

### *Search strategy*

Comprehensive electronic searches were conducted using Pubmed and Embase to look for articles up until 2019. A clinical librarian was consulted regarding the construction of the searches. Search terms used included "amyotrophic lateral sclerosis" or "ALS" or "motor neuron disease" or "MND" or "ALS/MND", combined with "telehealth" or "telemedicine" or "mhealth" or "ehealth" or, "telerehabilitation" or "telemonitoring" or "teleconsultation" or "digital technology" or "mobile technology" or "mobile app". Full search queries for Pubmed are shown in Supplementary material 1; we adjusted these for the other databases. Additionally, reference lists of identified articles were scrutinized and citations of these articles were checked using Google Scholar. Duplicates were removed using Mendeley software.

### *Inclusion criteria for review*

To be eligible, a study had to meet following criteria: (a) a full-text article with original research, (b) >75% of the study population had to be patients with ALS, (c) report on the use or implementation of telehealth in a healthcare setting, (d) published in English, (e) published in a peer-reviewed journal. Telehealth was defined as the provision of remote healthcare services through the use of digital and telecommunication technologies. The study methodology was assessed (i.e. study design and recruitment strategy), but was not an inclusion criterion for this review. Two reviewers screened titles and abstracts and selected relevant articles. A full-text assessment, also by two reviewers, determined which studies were eligible for inclusion based on the inclusion criteria.

### *Analysis method*

The framework for the qualitative data extraction was based on the Consolidated Framework for Implementation Research (CFIR) (9). This instrument specifies the determinants that affect the implementation of an innovation. The determinants in this review were divided into four domains from the CFIR: Innovation characteristics, patient/caregiver characteristics, healthcare professional characteristics, and the inner and outer setting. Innovation characteristics include the core components and complexity of the telehealth innovation that is being used or implemented. Patient/caregiver characteristics include the user-experiences/benefits, and compliance of patient/caregiver end-users. The healthcare professional characteristics include the user-experiences/benefits of healthcare professional end-users. The inner and outer setting includes the available resources, and finance and legislation that the organization

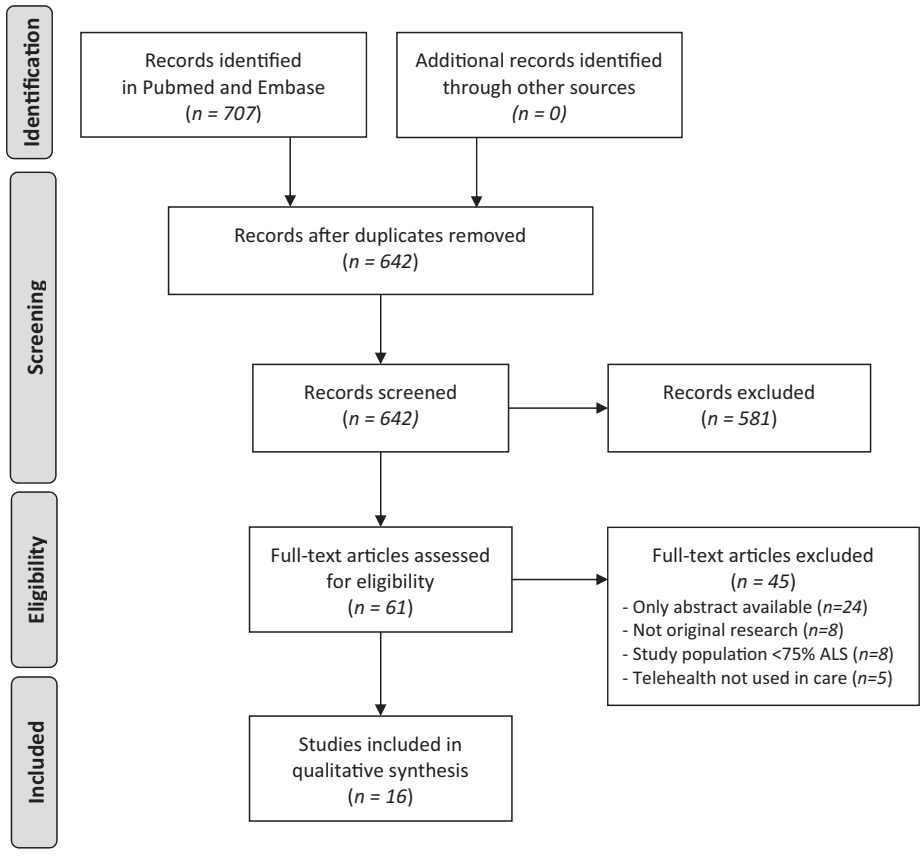


Figure 1. Literature selection flow diagram.

has to manage. The definitions of each determinant can be found in Table 1.

## Results

The final literature search was performed on the 6th of November 2019 and identified a total of 707 articles, 61 of which were reviewed for eligibility. After full-text analysis, 16 articles were included in the review. Reasons for exclusion are shown in Figure 1. At least 429 patients with ALS were included who used telehealth (one study did not report the number of patients); 65.9% of patients were male and the mean age was 60.5 years. Eight of sixteen studies reported the revised ALS Functional Rating Scale (ALSFRS-R) with a mean total score of 28.0; twelve of sixteen studies reported that 67.5% of patients were ventilated through either noninvasive ventilation (NIV) or tracheostomy invasive ventilation (TIV); and six of sixteen studies reported that 29.2% of patients had a gastrostomy. Details of the included studies can be found in Table 2.

Three main types of telehealth were identified: videoconferencing ($n = 5$), home-based self-monitoring ($n = 7$) and remote NIV monitoring ($n = 4$). Videoconferencing is any consultation between a patient/caregiver and a healthcare professional through real-time video. Home-based self-monitoring is the process in which patients (and their caregivers) manually perform measurements at home, and transmit data to the medical team and receive medical support via digital technology. Remote NIV monitoring is the process whereby a patient's digital NIV data are monitored remotely and transmitted to the medical team. One of the studies used the store and forward method in addition to videoconferencing. The store and forward method includes recording a patient assessment at home, after which the recording is stored and forwarded to the medical team for further assessment. The determinants of implementation per study can be found in Table 3 and an overview of all identified barriers and facilitators telehealth use/implementation is presented in Table 4.

### *Videoconferencing ($n = 5$)*

**Innovation characteristics.**

*Core components*. Studies reported that videoconferences were attended either by multiple healthcare professionals (10,11) or by one physician (12,13). In another study, during a home visit, a nurse set-up a videoconference for the patient and caregiver with the clinical director (14). A few studies required patients to login on the webserver with a personal ID code (10,12,13).

*Complexity*. One study reported issues with video and audio, but did not prevent any videoconferences from taking place (13). Audio issues

Table 2. Details of telehealth studies.

| Publication | Type of telehealth | Study design | Recruitment strategy | Study population | Characteristics of telehealth users (patients) |
|---|---|---|---|---|---|
| Geronimo et al. (10) | Videoconferencing | A pilot study investigating feasibility and acceptability. | A convenience sample consisting of patients who were deemed potentially eligible and would likely benefit from VC. | Telehealth ($N=20$) Caregiver ($N=20$) HCP ($N=15$) | Diagnosis (ALS), Age ($M=60$), Male (64%), ALSFRS-R ($M=24$) |
| Nijeweme-d'Hollosy et al. (12) | Videoconferencing | A pilot study investigating the effect of VC on the quality of care. | A convenience sample consisting of patients who liked working with computers. | Telehealth ($N=4$) | Diagnosis (ALS), Age ($M=42$), Male (75%) |
| van de Rijn et al. (13) | Videoconferencing | A retrospective chart review investigating the feasibility and utility of VC. | Patients who had at least one VC were included. | Telehealth ($N=97$) HCP ($N=5$) | Diagnosis (ALS), Age ($M=58$), Male (63%), V (57%), G (23%) |
| Selkirk et al. (11) | Videoconferencing | A retrospective cohort study investigating feasibility and effectivity. | Patients who received care for one year at the ALS center were included. Patients chose telemedicine based on preference, disability level or distance from the clinic. | Telehealth ($N=32$) Usual care ($N=36$) | Diagnosis (ALS), Age ($M=63$), Male (100%), V (72%), G (68%) |
| Pulley et al. (14) | Videoconferencing, Store and forward method | A pilot study investigating feasibility and acceptability. | Patients were included regardless of distance from the clinic, mobility or disease severity. | Telehealth ($N=18$) HCP ($N=7$) | Diagnosis (ALS), Age ($M=64$), Male (66%), ALSFRS-R ($M=25$) |
| Vitacca et al. (16) | Telephone-assisted self-monitoring | A pilot study investigating feasibility and patient/caregiver's satisfaction. | Patients with a caregiver were included. | Telehealth ($N=40$) Caregiver ($N=40$) | Diagnosis (ALS), Age ($M=63$), Male (60%), ALSFRS-R ($M=31$), V (78%), G (50%) |
| Vitacca et al. (17) | Telephone-assisted self-monitoring | A prospective study investigating feasibility and cost-effectiveness. | Patients who lived <80 km from the center were included. | Telehealth ($N=39$) Caregiver ($N=39$) HCP ($N=$ n.r.) | Diagnosis (ALS), Age ($M=62$), Male (54%), V (69%) |
| Vitacca et al. (18) | Telephone-assisted self-monitoring | A prospective study investigating nurse's utilization and costs. | Patients with an ALSFRS-R <40 were included. | Telehealth ($N=73$) | Diagnosis (ALS), Age ($M=61$), Male (60%), ALSFRS-R ($M=28$), V (49%), G (34%) |
| Paneroni et al. (19) | Telephone-assisted self-monitoring | A pilot study evaluating the feasibility of long-term self-monitoring. | Non-bulbar patients with a caregiver and an ALSFRS-R <35 were included. | Telehealth ($N=12$) | Diagnosis (ALS), Age ($M=53$), Male (75%), ALSFRS-R ($M=20$), V (58%) |
| Ando et al. (15) | App-based self-monitoring | A trial investigating the use and feasibility of telemonitoring. | Opportunity sampling was used to recruit patients. | Telehealth ($N=13$) | Diagnosis (MND), Age ($M=66$), Male (62%), ALSFRS-R ($M=22$), V (100%) |

(*Continued*)

Table 2. (*Continued*).

| Publication | Type of telehealth | Study design | Recruitment strategy | Study population | Characteristics of telehealth users (patients) |
|---|---|---|---|---|---|
| Ando et al. (20) | App-based self-monitoring | A qualitative study with semi-structured interviews on telehealth use. | Opportunity sampling was used to recruit patients who had completed a trial of telemonitoring. | Telehealth ($N=7$) Caregiver ($N=5$) | Diagnosis (MND), Age ($M=63$), Male (71%), V (100%) |
| Hobson et al. (21) | App-based self-monitoring | A randomized controlled pilot and feasibility study investigating the feasibility of an RCT. | Patients with ALS, PMA or PLS who showed a ≥2 point decrease on the ALSFRS-R in the last 18 months were included. | Telehealth ($N=20$) Usual care ($N=20$) Caregiver ($N=37$) HCP ($N=1$) | Diagnosis (ALS, PMA, PLS), Age (60), Male (70%), V/G (40%) |
| de Almeida et al. (22) | Remote NIV monitoring | An exploratory trial testing safety, acceptance, and accuracy of remote NIV monitoring. | Volunteering patients were included. | Telehealth ($N=$ n.r.) HCP ($N=9$) | Diagnosis (ALS), V (100%) |
| de Almeida et al. (23) | Remote NIV monitoring | A prospective, quasi-randomized controlled trial investigating costs and cost-effectiveness. | Patients who used a "bi-level" NIV device were screened and assigned into a group according to their residential area. | Telehealth ($N=19$) Usual care ($N=20$) | Diagnosis (ALS), Age ($M=60$), Male (70%), ALSFRS-R ($M=33$), V (100%), G (0%) |
| Tura et al. (24) | Remote NIV monitoring | A preliminary trial investigating feasibility. | n.r. | Telehealth ($N=15$) | Diagnosis (ALS), V (100%) |
| Pinto et al. (25) | Remote NIV monitoring | A prospective, quasi-randomized controlled trial investigating compliance, survival and healthcare utilization. | Patients who used a "bi-level" NIV device were assigned to one of two groups according to their residential area. | Telehealth ($N=20$) Usual care ($N=20$) | Diagnosis (ALS), Age ($M=60$), Male (70%), ALSFRS-R ($M=33$), V (100%), G (0%) |

ALSFRS-R: revised ALS functional rating scale; G: patients with gastrostomy; HCP: healthcare professional; M: Mean; NIV: noninvasive ventilation; n.r.: not reported; V: ventilated patients; VC: videoconference.

were solved by using a phone and a pager was used to troubleshoot technical issues. In one study, there were issues with buffering large video files, and an unstable and unreliable internet connection (14).

**Patient/caregiver characteristics.** *User-experiences and benefits.* Three studies reported that patients were satisfied with videoconferencing (12–14). One study reported that satisfaction with telehealth was not related to disease severity or travel distance (14). Reported benefits of patients included reduced travel burden, reduced clinical burden and time-saving (12,13). Remote consultation increased the continuity of care (11,13) and allowed more severely disabled patients to continue receiving specialist care (13). Caregivers reported a lack of physical evaluation by a healthcare professional (10).

*Compliance.* Several studies reported that patients felt comfortable and liked working with technology (10,12,14), which determined enthusiasm with telehealth (14). According to two studies, patients were willing to discuss most practical topics via remote consultation (e.g. medication, equipment, research, symptoms, and treatments) (12,13). However, one of these studies indicated reluctance of patients to discuss sensitive topics, such as acceptance/coping and end-of-life, during a remote consultation (12). These topics would require a face-to-face consultation. Patients were assisted by caregivers in order to use videoconferencing in some studies (10,12,13).

Table 3. Determinants of implementation categorized according to the CFIR domains.

| Publication | Innovation characteristics | Patient/caregiver characteristics | Healthcare professional characteristics | Inner and outer setting |
|---|---|---|---|---|
| *Videoconferencing* | | | | |
| Geronimo et al. (10) | Core components: Patients accessed VC through an internet browser using a unique ID code. The VC was attended by the patient, caregiver physician and nurse (other HCP's were optional). Telehealth was found to be feasible.<br>Complexity: Future changes to facilitate the use of VC include faster internet connection, compatibility with multiple types of hardware and internet browsers, direct eye contact through video, wider field of view and on-screen identification of HCP's. | User-experiences and benefits: Patients experienced reduced travel time and burden. The lack of physical evaluation was disliked by caregivers.<br>Compliance. Caregiver needed to assist patient. High comfort and familiarity with technology among patients. | User-experiences and benefits: HCP's judged telehealth as not equal to in-person care. HCP's lack the sense of touch. HCP's found communication and making recommendations adequate. A large portion of HCP's were unsatisfied with video and audio quality. | Available resources: The care protocol for telehealth was the same for in-clinic care, including similar staff requirement during consultations.<br>Finance and legislation: n.r. |
| Nijeweme-d'Hollosy et al. (12) | Core components: VC between patient, caregiver and physician. A web-application accessible through a virtual private network with a general ALS website and an tele-treatment environment, including planned consultation hours, a chat room and VC. External IT organization provided support.<br>Complexity: n.r. | User-experiences and benefits: Patients experienced reduced travel time. Patients were satisfied with telehealth and contact during VC.<br>Compliance: Patients were willing to discuss most topics through VC, except for acceptance and end-of-life. Patients liked working with computers and caregivers needed to assist when patients were too impaired. | User-experiences and benefits: n.r. | Available resources: A HCP provided an initial instruction session for patients.<br>Finance and legislation: n.r. |
| van de Rijn et al. (13) | Core components: Patients had to log into a virtual waiting room prior to VC. The VC was attended by the patient, caregiver and physician. HCP's used a pager for troubleshooting technical issues.<br>Complexity: All identified technical issues were related to video/ audio, none prevented VC from occurring. Audio issues were solved by using a phone. | User-experiences and benefits: It was less necessary to travel for consultations. Patients experienced time saving and a reduction in clinic burden. More continuity of care was seen in patients in a later phase of disease.<br>Compliance: Patients were satisfied with VC. Majority of patients were assisted by caregiver. Patients in every phase of the disease preferred VC. Patients were willing to discuss most topics through VC. | User-experiences and benefits: HCP's experienced easier communication with local therapist. HCP's are uncertain about proper assessment and lack a physical examination. No time saving for HCP's. Most topics could be discussed via VC. | Available resources: A team of coordinators and engineers of the telehealth division provided training and technical support to physicians and patients. Finance and legislation: There was a lack of reimbursement for telehealth. |
| Selkirk et al. (11) | Core components: Prior to a VC patients were evaluated by their local team, a nurse called patient to assess burden, QoL and ALSFRS-R-R, and patients' needs were determined during a bi-weekly meeting of the medical team. The VC was attended by the patient, caregiver, local care team, nurse and neurologist (other HCP's were optional).<br>Complexity: n.r. | User-experiences and benefits: VC allows for more continuity of care throughout the disease. It was less necessary to travel for consultations. Quality of care was equivalent between regular care and telehealth.<br>Compliance: n.r. | User-experiences and benefits: n.r. | Available resources: The care protocol for telehealth was the same for in-clinic care, including similar staff requirement during consultations.<br>Finance and legislation: n.r. |

(*Continued*)

Table 3. (*Continued*).

| Publication | Innovation characteristics | Patient/caregiver characteristics | Healthcare professional characteristics | Inner and outer setting |
|---|---|---|---|---|
| Pulley et al. (14) | Core components: A home visit by a nurse for video recording of clinical assessments. Video of the assessments was cut and assessed by HCP's. Reports by the HCP's were sent to clinical director who created a care plan. The plan was conveyed through VC. The VC was attended by the patient, caregiver, clinical director and at-home nurse.<br>Complexity: The lack of availability of patient and nurse delayed VC. Buffering of large video files was a major issue. Internet connection was often unstable and unpredictable. | User-experiences and benefits: It was less necessary to travel for consultations.<br>Compliance: Patients were satisfied with telehealth, independent of ALS severity and distance from center. Comfort with using technology determines enthusiasm. | User-experiences and benefits: Mixed experiences with the ease of the process, lack of physical contact and time requirement. HCP's had limited availability on clinic days. The information obtained from telehealth was sufficient. HCP's assessed the video at their convenience. | Available resources: All member of the multidisciplinary team provided extensive training to the nurse in assessment skills. Clinic director conducted videoconferences with nurse at patient's home. Inexpensive commercial devices were used.<br>Finance and legislation: There was a lack of reimbursement for telehealth. There was no team conference to discuss a case. |
| *Home-based self-monitoring* | | | | |
| Vitacca et al. (16) | Core components: Patients performed daily at-home assessments of pulse oximetry. Weekly scheduled call from a nurse for a clinical interview, consultation planning, updating clinical data (oximetry) and redirecting to a general practitioner or specialist. There was a 24h availability of second-opinion by a respiratory therapist. A call-center facilitated communication.<br>Complexity: n.r. | User-experiences and benefits: Patients were extremely satisfied with the nurse assistance. Patients had more confidence in handling the disease. 24h availability of second-opinion was valuable. Telehealth provided autonomy and continuity of information and care.<br>Compliance: Majority of patients were assisted by caregiver. Caregiver spoke/assisted when patient lost speech. Devices were simple to use. | User-experiences and benefits: n.r. | Available resources: Nurse monitored 50 patients with a clinical ALS-card. On demand availability of a nurse during working hours. Providing 24/7 call-center service and second opinion by a general practitioner or specialist. No dedicated therapist needed for second opinion.<br>Finance and legislation: System is believed to be sustainable in terms of cost and staff time required (no analysis conducted). Variety of fixed and variable costs. |
| Vitacca et al. (17) | Core components: Patients performed at-home assessments of MAC, pulse oximetry and airway suctioning. MI-E was provided for patients in which blood oxygen saturation could not be restored. Weekly scheduled call from respiratory therapist. Calls from patients were redirected by a nurse to one of the specialists for consultation or home-visit. A call-center facilitated communication.<br>Complexity: n.r. | User-experiences and benefits: Fewer hospital admissions and emergency room visits. Patients experienced an increased feeling of security for home management and considered the intervention effective.<br>Compliance: n.r. | User-experiences and benefits: HCP's experienced an increased feeling of security for home management. | Available resources: On demand telephone access to a triage nurse who directed calls to a specialist. Therapist called patients weekly. Patients were trained to restore blood oxygen saturation through assisted coughing and emergency room visits.<br>Finance and legislation: There was a lack of reimbursement for telehealth. Additional costs were home-visits and device rental. Intervention was shown to be cost-effective, due to reduced hospitalization costs. |

(*Continued*)

Table 3. (*Continued*).

| Publication | Innovation characteristics | Patient/caregiver characteristics | Healthcare professional characteristics | Inner and outer setting |
|---|---|---|---|---|
| Vitacca et al. (18) | Core components: Patients performed daily at-home assessments of pulse oximetry. Weekly scheduled calls from a nurse for a clinical interview, updating clinical data (oximetry) and redirecting to a general practitioner or specialist. In addition, patients could request calls 24/7. A call-center facilitated communication. An ALS card-of-risk guided telephone-accessed clinical interviews and facilitated monitoring, care and interoperability.<br>Complexity: n.r. | User-experiences and benefits: n.r.<br>Compliance: Caregiver needed to assist patients. | User-experiences and benefits: n.r. | Available resources: Providing on demand access to a nurse during working hours and 24/7 call-center service and second opinion by therapist. Providing nurses with training to use the ALS-card to monitor disease status and to guide clinical interviews. Clinical ALS-card saved time and expenses on telephone calls.<br>Finance and legislation: Health expenditure policy needs optimization of staff's costs. Large variety of fixed and variable costs. Costs were extrapolated on long-term steady-state telehealth activity, including other chronic diseases. |
| Paneroni et al. (19) | Core components: Patients performed daily assessments of PCEF, pulse oximetry and respiratory discomfort. Patients also reported changes in respiratory and clinical condition in a diary. Bi-weekly scheduled telephonic support from a physiotherapist. Data was mostly transmitted through email or telephone.<br>Complexity: High number of daily assessments and complex of reporting. | User-experiences and benefits: n.r.<br>Compliance: Caregivers needed to assist when a patient was too impaired. Patients showed low adherence with the monitoring protocol. | User-experiences and benefits: n.r. | Available resources: Provision of telephonic support by a dedicated therapist.<br>Finance and legislation: n.r. |
| Ando et al. (15) | Core components: A tablet-style device was used by patients for answering questions and transferring nocturnal pulse oximetry data weekly. Patients could message the clinical team at any time. All patients received education on symptoms related to chest infections and respiratory management<br>Complexity: n.r. | User-experiences and benefits:<br>Compliance: Patients showed high levels of adherence with telemonitoring. | User-experiences and benefits:<br>HCP's had the ability to remotely monitor patient's symptoms effectively and offer timely and appropriate support. | Available resources: n.r.<br>Finance and legislation: n.r. |
| Ando et al. (20) | Core components: A tablet-style device that allows clinicians to monitor patient outside of the clinic, regarding their symptom changes, NIV related related issues, nocturnal blood oxygen saturation levels, and patient-ventilator interaction data. The system generated alerts for symptom worsening. | User-experiences and benefits: Patients experienced timely interventions, which resulted in improved physical and psychological well-being. Reduced number of (unnecessary) clinic visits, saving time and costs. Increased self-awareness was experienced by patients.<br>Compliance: Patients were supportive of | User-experiences and benefits:n.r. | Available resources: n.r.<br>Finance and legislation: n.r. |

(*Continued*)

Table 3. (*Continued*).

| Publication | Innovation characteristics | Patient/caregiver characteristics | Healthcare professional characteristics | Inner and outer setting |
|---|---|---|---|---|
| | Complexity: There were some technical issues, but these were often minor and quickly resolved. The message function lead to frustrations due to too small keys and short time allowance for formulating a message. | regular monitoring and generally experienced the use of technology as easy. Using the message function was frustrating and challenging for some patients. | | |
| Hobson et al. (21) | Core components: A user-centered telehealth service including weekly assessments of body weight and balance, and using a tablet to complete questions on functional ability, progression, symptoms, well-being. A nurse could view the data on a website; she phoned patients, expedited appointments and lialised the medical team. The system generated alerts for disease worsening. Complexity: There were some technical issues, but these were resolved. The connectivity of wifi-enabled scales was unreliable. The software was user-friendly and accessible for patients/caregivers. | User-experiences and benefits: Patients/caregivers experienced improved awareness of the disease, reassurance due to increased monitoring and a better connection with the HCP's. Face-to-face technology training was perceived as important. Patients/caregivers would recommend telehealth to others. Compliance: Good adherence to self-monitoring was observed. Accordingly, self-monitoring was easy, not tiring or time consuming, and not distressing. Some experienced difficulty using technology due to upper limb disability. Caregivers assisted when patients were too impaired. Low perceived ability to master technology, due to inexperience with technology. | User-experiences and benefits: Telehealth use was very easy and cost little time. The nurse could identify early problems, however the information from monitoring was sometimes not detailed enough. Repetitive alerts were frustrating for the nurse and required more time. On most occasions the information from monitoring was sufficient for a HCP to make appropriate decisions. | Available resources: Provision of training for technology use and provision of technical support. Finance and legislation: n.r. |
| *Remote NIV monitoring* | | | | |
| de Almeida et al. (22) | Core components: An NIV device with flexible use of electronic slots and bi-directional data transmission was used for remote monitoring.A helpline was available for technical issues or worsening of clinical condition. Technician monitored NIV data and flagged physician for immediate change of settings. Complexity: The system was suitable and worked well. The connection was found to be robust for transmission of data and setting changes. The speed of data extraction was a limitation. There was need of a fixed telephone line. There were issues with confidentiality. | User-experiences and benefits: Reduced need to travel to adjust NIV settings. Compliance: Positive comments from patients on easiness of setting arrangements. Automatic monitoring limited the need for manual intervention. | User-experiences and benefits: HCP's lacked the sense of touch. The system eased communication with patient. HCP's had the ability to remotely monitor and change NIV settings. | Available resources: Helpline support was provided. Testing phase was required to test safety, acceptability and accuracy. Hiring technician for monitoring of NIV data and checking the procedure and mistakes. Finance and legislation: Unresolved issues with licensure, reimbursement, telecommunications infrastructure and robust analysis of cost-effectiveness. |

(*Continued*)

Table 3. (*Continued*).

| Publication | Innovation characteristics | Patient/caregiver characteristics | Healthcare professional characteristics | Inner and outer setting |
|---|---|---|---|---|
| de Almeida et al. (23) | Core components: An NIV device with bi-directional data transmission was used for remote monitoring.<br>Complexity: n.r. | User-experiences and benefits: Improved patient enablement and confidence with handling disease. Reduced need to travel to adjust NIV settings. Fewer office visits and hospital admissions.<br>Compliance: Automatic monitoring limited the need for manual intervention. | User-experiences and benefits: HCP's had the ability to remotely monitor and change NIV settings. | Available resources: Higher number of setting changes at start, but lower over the entire period. Fewer office visits and hospital admissions.<br>Finance and legislation: Big initial investment needed, but cost effective over time. Device rent was higher in remote monitoring group. |
| Tura et al. (24) | Core components: An NIV device with flexible use of electronic slots was used for remote monitoring. The wireless connection was found to be robust. A general practitioner or specialist could see data on the web application and send a (voice) message to the patient.<br>Complexity: The device was easy to handle, with additional remote control and connection to a smartphone, personal digital assistant or notebook. | User-experiences and benefits: Reduced need to travel to adjust NIV settings.<br>Compliance: Patients reported devices as easy to use, and device settings were easy to adjust. The remote control was appreciated. Automatic monitoring limited the need for manual intervention. | User-experiences and benefits: HCP's had the ability to remotely monitor and change NIV settings. | Available resources: n.r.<br>Finance and legislation: Lack of information on cost-effectiveness. |
| Pinto et al. (25) | Core components: An NIV device with bi-directional data transmission was used for remote monitoring. A helpline was available for NIV compliance follow-up, setting changes or medical advice. Technician monitored NIV data and flagged physician for immediate change of settings. The NIV device had bi-directional data transmission.<br>Complexity: The system was suitable and worked well. The speed of data extraction was limited. There was a need of a fixed telephone line. | User-experiences and benefits: Fewer hospital admissions. Reduced need to travel to adjust NIV settings.<br>Compliance: Automatic monitoring limited the need for manual intervention. | User-experiences and benefits: HCP's had the ability to remotely monitor and change NIV settings. | Available resources: Providing helpline support. Testing phase was required to test safety, acceptability and accuracy. Hiring technician for monitoring of NIV data. Higher number of NIV setting changes at start, but 50% lower over the entire period.<br>Finance and legislation: Reduced hospitalization costs, due to fewer hospital admissions. |

ALSFRS-R: revised ALS functional rating scale; HCP: healthcare professional; MAC: mechanical assisted coughing; MI-E: mechanical in-exsufflation; NIV: noninvasive ventilation; n.r.: not reported; PCEF: peak cough expiration flow; QoL: quality of life; VC: videoconference.

Table 4. The barriers to and facilitators of telehealth use/implementation in ALS care.

| | |
|---|---|
| *Facilitators* | |
| Innovation characteristics | Complexity<br>• Robust wireless connection<br>• User-friendly devices<br>• Additional aids for devices |
| Patient/caregiver characteristics | User-experiences and benefits<br>• Reduced travel time and burden<br>• Reduced clinic burden<br>• Fewer hospital admissions/ emergency room visits<br>• Increased feeling of enablement/ self-confidence<br>• Increased feeling of security<br>Compliance<br>• Easy to use devices<br>• Comfort and familiarity with using technology<br>• Caregiver assistance<br>• Automatic monitoring<br>• High adherence to self-monitoring |
| Healthcare professional characteristics | User-experiences and benefits<br>• Increased feeling of security<br>• Insight into remote monitoring of data<br>• Better communication |
| Inner and outer setting | Available resources<br>• Ongoing support for end-users<br>• Training of end-users<br>• Standardized clinical assessment (card-of-risk)<br>Finance and legislation<br>• Sustainable in costs and time requirement<br>• Cost-effective MI-E rental/ NIV use<br>• Inexpensive commercial devices<br>• Reduced hospitalization costs |
| *Barriers* | |
| Innovation characteristics | Complexity<br>• Cumbersome monitoring protocol<br>• Technical issues<br>• Slow internet connection<br>• Slow data extraction/ buffering |
| Patient/caregiver characteristics | User-experiences and drawbacks<br>• Lack of physical evaluation/contact<br>Compliance<br>• Low adherence to self-monitoring<br>• Unwilling to discuss sensitive topics through telehealth |
| Healthcare professional characteristics | User-experiences and benefits<br>• Lack of physical evaluation/ contact<br>• Uncertainty about comprehensive medical assessment<br>• No time saving/ costing extra time |
| Inner and outer setting | Finance and legislation<br>• Big initial investment<br>• Large variety of fixed and variable costs<br>• Lack of reimbursement<br>• Lack of cost-effectiveness analyses |

ALSFRS-R: ALS functional rating scale; HCP: healthcare professional; G: patients with gastrostomy; M: Mean; MAC: mechanical assisted coughing; MI-E: mechanical in-exsufflation; NIV: noninvasive ventilation; n.r.: not reported; PCEF: peak cough expiration flow; QoL: quality of life; V: ventilated patients; VC: videoconference.

**Healthcare professional characteristics.**

*User-experiences and benefits.* One study showed that healthcare professionals were generally satisfied with the communication and provision of care during videoconferences (10). In addition, videoconferencing enabled a local therapist to attend the consultations, which normally was not possible with in-clinic care (12). The healthcare professionals in two studies were able to discuss most, but not all, topics (as reported by patients) through videoconferencing (12,13). Despite these positive experiences, several studies indicated the lack of a sense of touch perceived by healthcare professionals (10,13,14) and one study reported that healthcare professionals might be uncertain about whether videoconferencing allows for an appropriate medical assessment (13). Additionally, in one study, healthcare professionals expressed dissatisfaction with the quality of the video and audio, and reported that telehealth was not equal to in-person care (10). One study reported mixed opinions on time requirement and ease of the process of the store and forward method (14).

**The inner and outer setting.**

*Available resources.* Studies showed that the care protocol for videoconferences was the same as for in-clinic care, including equal staff requirement during consultations (10,11). In a couple of studies, training of healthcare professionals and patients was required for using videoconferencing (12–14). Technical support for healthcare professionals in two studies was provided by an external information technology organization (12) or the internal telehealth division (13).

*Finance and legislation.* One study reported a lack of reimbursement for telehealth (13).

*Home-based self-monitoring (n = 7)*

**Innovation characteristics.**

*Core components.* Studies included the at-home assessment of oximetry (15–20), questions on functional status and symptoms (15,20,21), manually or mechanically assisted coughing, airway suctioning and mechanical in-exsufflation (MI-E) (16), peak cough expiration flow, respiratory discomfort and a clinical diary on changes in clinical condition (19), and body weight and balance (21). Some studies used a monitoring protocol with daily assessments (17–19), while a number of other studies applied a weekly or bi-weekly monitoring protocol (15,20,21). Self-monitored data was transmitted either through telephone or a tablet device (15–21).

*Complexity.* One study reported that self-monitoring seemed to be too cumbersome due to the large number of daily assessments and complexity of reporting (19). In contrast, adhering to the self-monitoring protocol in other studies was considered to be easy, due to an appropriate frequency of monitoring and user-friendly technology (15,21). Specifically oximeters and tablet devices were reported to be user-friendly (17,20,21). In some cases, the self-monitoring protocol was reinforced by home visits (17) or telephone calls (17,21).

**Patient characteristics.**

*User-experiences and benefits.* Satisfaction with telehealth was reported in three studies (15,17,21). Patients reported increased awareness of the disease (15,21), more confidence in dealing with the disease (17) and an increased feeling of security for home management of respiratory symptoms (16). A reduction in hospital admissions was seen in one study (16) and in another study, the number of unnecessary clinic visits was reduced, resulting in a saving in time and costs (15).

*Compliance.* In several studies, patients were assisted by a caregiver when they were too impaired (16–18,21). Patients reported in two studies that devices worked well and were easy to use (15,17), but patients in another study had difficulty using a tablet device due to upper limb disability (21). One study reported that patients showed low compliance with a monitoring protocol with multiple daily measurements (19). In contrast, two studies reported high adherence with a (bi-)weekly monitoring protocol (15,21). Accordingly, patients and caregivers in these studies reported that self-monitoring was easy, not time-consuming nor tiring.

**Healthcare professional characteristics.**

*User-experiences and benefits.* It was reported that telehealth allowed healthcare professionals to monitor symptoms effectively, provide timely support (15) and make appropriate decisions in care (21). One study reported that telephone-assisted self-monitoring increased healthcare professionals' feeling of security for home management of respiratory symptoms (16). A telehealth nurse reported that telehealth was easy to use and not time-consuming (21). However, she also reported that information from self-monitoring was often not detailed enough and that repetitive alerts lead to frustration.

**The inner and outer setting.**

*Available resources.* Studies reported that medical support could be requested through a telephone-call (15–21), a message system (15,20) or email (21), and that support was provided by either a therapist or a nurse. In one study a telehealth nurse remotely monitored patients through a clinical portal, phoned patients, expedited appointments and liaised the medical team (21). In another study, nurses used a standardized ALS card-of-risk to guide telephonic clinical interviews (18). Nurses in these studies required training for operating the ALS card-of-risk and the clinical portal. Healthcare professionals provided face-to-face training to patients in the process of restoring blood oxygen saturation (16) and in operating a tablet (21).

*Finance and legislation.* It was reported that on-demand MI-E rental was cost-effective compared to continuous rental and that fewer hospital admissions reduced hospitalization costs (16). Telephone-assistance was believed to be sustainable in terms of cost and staff requirement (17). Reported barriers to the continuation of telehealth use were a lack of information on cost-effectiveness (18) and a lack of reimbursement (16). A large variety of fixed and variable costs related to tele-assistance were seen in two studies (17,18).

*Remote NIV monitoring (n = 4)*

**Innovation characteristics.** *Core components.* Studies reported using an NIV device with bi-directional data transmission, which allowed for automatic transmission of NIV data and remote adjustment of NIV settings (22–25). NIV devices had flexible use of electronic slots, which allowed arrangements to be tailored to patients' needs (22,24). In two studies, patients had access to a helpline for on-demand medical or technical support; technicians monitored NIV data and flagged the physician to immediately change settings (22,25).

*Complexity.* Two studies reported that the NIV devices were easy to use and showed robust wireless connection tests for transmission of data and setting changes (22,24). In one study, additional functionalities and aids were provided to facilitate NIV use (24). The speed of data extraction was limited in two studies (22,25).

**Patient characteristics.** *User-experiences and benefits.* The main benefit for patients was a reduced need to travel to the clinic for adjustment of NIV settings (22–25). Additionally, fewer hospital admissions were reported (23,25). In one study, patients experienced improved enablement and more confidence in managing the disease (23).

*Compliance.* The bi-directional and automatic functionality limited the need for manual intervention by patients and caregivers for monitoring. Patients reported that the NIV devices were easy to use, and that settings were easy to change/arrange (22,24). In one study, patients appreciated the extra aids that were provided to facilitate NIV use (24).

**Healthcare professional characteristics.** *User-experiences and benefits.* One study reported that remote monitoring facilitated communication with the patient, but that healthcare professionals missed the sense of touch.(10) In all studies, healthcare professionals had the ability to remotely monitor NIV and adjust settings, which would not be possible with usual in-clinic care (22–25).

**The inner and outer setting.** *Available resources.* In some studies, on-demand support was provided through a helpline (22,25). A testing phase was required in two studies and (cardio-pulmonology) technicians were hired to monitor NIV data (22,25) and check procedures and mistakes (22). Two studies reported that the number of NIV setting changes was 50% lower over the entire period of NIV use, compared to usual care, hence saving time (22,25).

*Finance and legislation.* One study reported that a large initial investment is required to set up telehealth and that remote NIV monitoring in ALS care is cost-effective (23). Another study reported that the number of hospital admissions was reduced, which resulted in lower hospitalization costs (25). Reported barriers to telehealth implementation were a lack of robust cost-effectiveness-analysis (22,24) and issues with reimbursement (22).

## Discussion

This review identified three different types of telehealth used in ALS care and showed that telehealth was mainly targeted at patients with respiratory impairment. Furthermore, we found that the barriers and facilitators of telehealth implementation in ALS care were consistent with the determinants identified in other healthcare settings. The main barriers hindering implementation of telehealth in ALS care were related to issues with finance and legislation, and lack of personal contact perceived by healthcare professionals.

It was noticeable in this review that the proportion of patients who were ventilated through NIV or TIV (68%) was much higher compared to the general ALS population (18–36%) (26,27). In addition, 10 of 11 remote monitoring studies focused primarily on respiratory function, such as oximetry, (assisted) coughing, respiratory symptoms, and NIV. These findings demonstrate that patients in the included studies are not representative of a general ALS population and that telehealth is focused on respiratory function up until now.

### *Determinants of implementation*

Our results indicate that patients with ALS (and their caregivers) have a positive attitude toward the use of telehealth. This may be attributed to patients' perceived benefits of telehealth (e.g. an increased feeling of enablement, reduced travel and clinical burden) and good compliance to telehealth use (i.e. easy to use devices, comfort with using technology and caregiver assistance). A positive attitude of patients/caregivers is a facilitator for implementation as it increases acceptance of telehealth and positively influences the attitude of healthcare professionals (28,29). It should be noted that several studies recruited a convenience sample of patients, who were likely to benefit from telehealth, or liked working with technology. This may have affected patients' experiences. Results suggest that healthcare professionals in ALS care have a more negative attitude toward telehealth. Despite being positive about communication through telehealth, healthcare professionals mostly reported barriers, such as technical issues, a lack of

physical evaluation/contact and issues with a comprehensive medical assessment. A negative attitude among healthcare professionals creates resistance to telehealth and is a known barrier to implementation (28,29). Regrettably, more than half of the studies did not evaluate user-experiences of healthcare professionals and therefore lack this information. Two important facilitators in this review that positively influenced the attitude of end-users were the provision of training and ongoing support. Despite requiring more staff time, the provision of training and ongoing support ensures that end-users are able to apply technology properly, which is essential for a successful implementation (28,29). One of the main barriers to implementation of telehealth in ALS care was issues related to finance and legislation. Studies mostly reported a lack of robust cost-effectiveness analyses and a lack of reimbursement for telehealth. These are important issues that are also known in other healthcare settings, and hinder the implementation and integration of telehealth in ALS care. There was, however, evidence to support that on-demand MI-E rental and remote NIV monitoring were cost-effective, primarily due to a lower number of emergency room visits and hospital admissions. This financial benefit of telehealth is a facilitator for implementation and should be investigated in future research.

### *Clinical implications*

Current telehealth innovations are mainly targeted at a subgroup of patients with ALS, which means that a substantial portion of the ALS population does not benefit from them. Ideally, all patients with ALS should be able to benefit from the use of telehealth, irrespective of the disease stage or type of impairments. Recent evidence has shown that this is feasible and of benefit for patients with motor neuron disease (21). For this reason, future innovations should remotely monitor all relevant domains of functioning from early on in the disease. This will help healthcare professionals with (a) detecting early signs and symptoms, (b) informing patients about changes in all aspects of the disease and (c) providing timely care and information. Additionally, if started in an early disease stage, patients will be able to receive training and become familiar with technology and remote monitoring before becoming severely impaired.

To make sure a telehealth innovation truly meets the needs of end-users, both patients and healthcare professionals should be involved throughout the development process (21,30). The involvement of end-users will promote a positive attitude toward telehealth and increase acceptance, thus facilitating implementation (28,29). Furthermore, telehealth innovations should be personalized, as the rate of disease progression is highly variable and the care needs of patients with ALS are ever-changing. The personalization of telehealth promotes patient engagement (31) and involves the tailoring of monitoring frequency, clinic visit scheduling and the provision of care and information. To further increase patient engagement with remote monitoring, notifications and personal feedback could be provided (31).

To improve remote monitoring and the usability of monitoring-data for research purposes, standardized outcome measures should be established and patients should be involved in determining which measures are relevant. Ideally, outcome measures should be associated with disease progression and survival, as this will help with the timely provision of interventions, assistive devices, and information. Examples of such outcome measures are the ALSFRS-R, weight loss and vital capacity (32–34). Also, assessments on cognition, quality of life and caregiver burden could be included to facilitate psychological support.

### *Future research*

In order to improve the implementation of telehealth in ALS care, future studies should be aimed at identifying the determinants of implementation and investigating how they affect the success of telehealth. Improved reporting (on determinants) will help to create a more detailed overview of relevant determinants, which is essential to guide healthcare professionals in the implementation of future telehealth innovations in ALS care. Furthermore, future research should focus on investigating the cost-effectiveness. Robust analyses will specifically facilitate the use of telehealth beyond the initial pilot phase.

### *Limitations*

The main limitation of this review is that there were no studies primarily aimed at identifying the determinants of implementation. As a result, positive and negative aspects of telehealth might not have been reported or were not specifically reported as barriers or facilitators of telehealth implementation. For this reason, we may have missed a number of potential barriers, such as issues with (national) policy and incompatibility with the current infrastructure. Another limitation is that a number of studies included a convenience sample, which may have resulted in biased patients' experiences.

## Conclusion

Our findings show that telehealth in ALS care is well-received by patients and their caregivers, as a result of user-friendly technology and experienced benefits. The provision of training and ongoing support to end-users has shown to be key for a successful telehealth implementation. Issues with

reimbursement of telehealth and lacking information on cost-effectiveness were the main challenges. Future research should specifically focus on reporting barriers and facilitators to guide future telehealth implementation and help design new implementation strategies.

## Declaration of interest

The authors declare no conflicts of interest and that the current study is their own work.

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