Telehealth in patients with ALS: A systematic review assessing the barriers and facilitators for implementation

# Abstract

**Introduction:** Patients suffering from Amyotrophic Lateral Sclerosis have a life expectancy of 2-4 years after diagnosis. No cure is available for them. Telehealth can decrease the burden of care for patients as well as caregivers. In this review we wish to determine the current use of telehealth and if barriers and facilitators for implementation of telehealth solutions have changed in ALS care since a previously published review, and consequently also since the start of the COVID-19 pandemic

**Methods:** Articles were searched on PubMed and screened for eligibility. Telehealth use in ALS care was described and reported barriers and facilitators for implementation were summarized and structured according to the CFIR framework.

**Results:** A total of 18 articles were included. Two types of telehealth used in ALS care were distinguished. Telehealth on the basis of remote consultations and remote monitoring of patients via monitoring platforms. Teleconsultations could be done either via video- or telephone consultations. Monitoring platforms could incorporate questionnaires or could use assistive equipment. Facilitators were found in the relative advantage category as well as the individual characteristics. Barriers were found mostly in the outer setting

**Conclusions:** Telehealth is more widely used in ALS care than before, to which the COVID-19 pandemic might be partially responsible. Telehealth in ALS care is done via remote consultations and via monitoring platforms. A facilitator for the implementation of telehealth is the decreased burden of care for both patients and caregivers. Barriers are mostly related to regulations and reimbursement strategies are lacking. Remote monitoring of patients has increased and is perceived as beneficial by both clinicians and patients.

# Layman’s summary

Care for patients with chronic diseases can be burdensome to the patient. Long days in the hospital with many different appointments are considered tiring and are often planned periodically. Telehealth is a method that enables patients to receive (part of) their care from home, either via remote consultations or via remote monitoring. A review about the telehealth care of patients suffering from ALS and the barriers and facilitators associated with implementation of this care was conducted in 2018. We wish to determine if the care as well as the barriers and facilitators associated with implementation has/have changed. In this systematic review we included all articles published between 2018 and oktober 2022 concerning these topics, and extracted the barriers and facilitators. These were placed in the CFIR framework for implementation.

Use of telehealth is more widespread than in 2018, when patients receiving ventilation benefited most. All patients can now potentially benefit from telehealth. Telehealth is, like in 2018, given in two different ways. Via remote consultations and via remote monitoring. While some experimentations with remote monitoring are starting up, for example remote monitoring of diet, no big new ways of remote monitoring are implemented. Facilitators are mostly seen in the individual characteristics category and the relative advantage construct. Patients find that telehealth decreases disease- and care burden. Fewer barriers than before concern the healthcare professionals. The main barriers are seen in the outer setting, with problems concerning legislation and insurance reimbursement. In clinical trials a lot of new ways of monitoring the patient are being implemented. These innovative monitoring strategies will surely make their way to the care of the patient in the near future.

# Introduction

Amyotrophic Lateral Sclerosis (ALS) is a devastating neurodegenerative disease characterized by the loss of upper- and lower motor neurons.1 This leads to progressive muscle weakness, paralysis and eventually death by respiratory muscle failure. Location of disease onset could be spinal or bulbar, and life expectancy is 2-5 years after initial diagnosis. Initial presentation, disease progression, and life expectancy all vary greatly between patients.2 No cure has been found for ALS yet. Riluzole currently is the only approved drug increasing tracheostomy-free life expectancy with months.3 ALS care is organised within a large multidisciplinary team focussing on alleviation of symptoms and increasing quality of life.4,5

Because of the heterogeneous clinical picture and increasing disability of patients, ALS patients have ever-changing needs in their care. To ensure that the patient receives the proper care, close monitoring of the patients course of disease is necessary, but the burden on patients as well as caregivers to adhere to monitoring through hospital visits increases with disease progression and subsequent disabling of patients. Having less monitoring appointments is no option, because important parts of ALS care, including ventilatory support and other supportive interventions, should be started in time to keep the quality of life of the patient as high as possible.4

One emerging solution for this is Telehealth. Telehealth or Telemedicine is a rapidly progressing field that aims at providing digital health services to patients.6 It explores the possibilities of facilitating care for a patient from home, using i.e. videoconferencing, chat functions, data registration via sensors, and periodic questionnaires. In less developed regions telehealth might also constitute consultations conducted over telephone.7,8 Before COVID-19, an estimated 46% of care provided in hospital could be provided at home.9 Several studies ran pilots to implement telehealth strategies in patient groups suffering from ALS or other neurodegenerative diseases, but many of these studies did not make it out of the pilot phase. A systematic review of these studies has been published right before the COVID-19 pandemic hit. This study identified three main types of telehealth: Video conferencing, home-based self-monitoring, and remote NIV monitoring. The study concluded that patients and caregivers have a generally positive mindset towards telehealth, while healthcare professionals show mixed reactions, perceiving more possible barriers for implementation**.**10During the pandemic, many healthcare institutions were forced to implement some forms of telehealth, because it was not deemed responsible for patients to visit to hospital, and nearly all regular care was scaled down.7,8,11–13Healthcare professionals, as well as patients, were forced to work with telehealth over a longer period of time. Since this period forced all parties to work with telehealth solutions, it is likely that feelings of resistance have decreased, as professionals and patients will be grateful for any sort of contact during the pandemic.

This systematic review summarizes the available evidence on the current use of telehealth in ALS Care and the barriers and facilitators experienced by patient, caregiver or healthcare professionals concerning telehealth implementation in ALS care.10

# Methods

**This review is an update of a systematic review addressing barriers and facilitators that influence implementation of telehealth in ALS care that were reported in studies published prior to November 2018.10**

**Search strategy**

**An extensive literature search was curated and conducted on Pubmed on 10-10-2022. The search included terms (MesH and text words) for the diagnosis motor neuron disease in combination (through ‘AND’) with search terms on telehealth. The extensive search strategy is outlined in appendix 1.**

**Inclusion criteria**

As a general inclusion criterion in this first step, all original, English publications that describe any digital health intervention in the care for patients with MND such as mobile technology, web-based applications, or digital communication tools used were included.

Articles not meeting this criterion were excluded. After a first selection was made based on title and abstract, full text review was done. To be included in the analysis, studies were selected to meet the following criteria:

* The study population must consist of at least 50% ALS patients.
* The article must report on the development of a system for remote ALS/MND care.
* The article must report about at least one of the following:
  + The satisfaction of ALS/MND patients and/or (family)caregivers on the use of digital technology in remote ALS/MND-care.
  + The barriers and/or facilitators for the implementation or adoption of the implementation or adoption of digital health services in motor neuron disease care.

Studies were excluded if 1) They were included in the previous systematic review.10; 2) They were abstracts, editorials, or conference proceedings; or 3) Focused only on the technical aspects of digital technology rather than experiences in implementation.

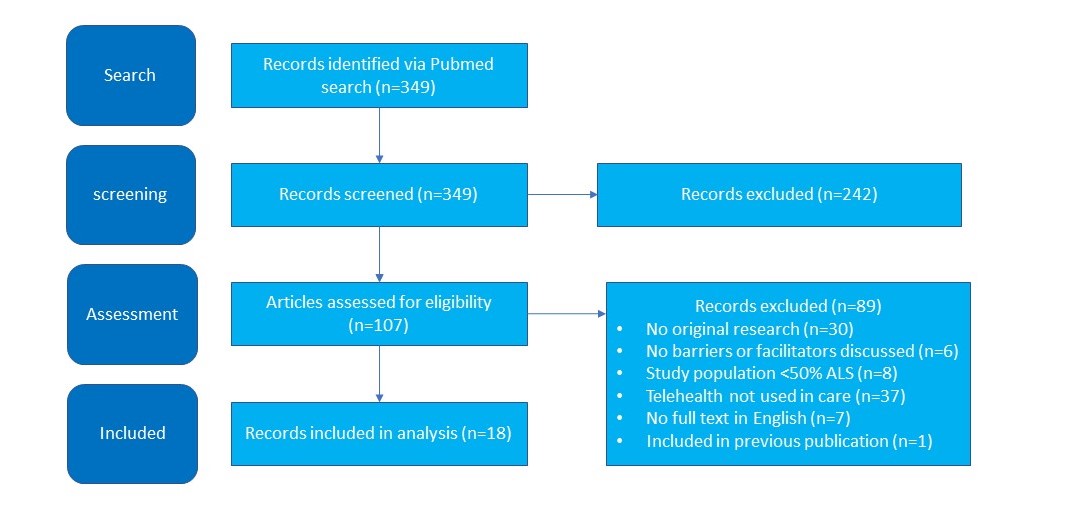
**Data extraction**

During full-text review of articles, data were extracted using a data collection form that was based on the Consolidated Framework for Implementation Research (CFIR). This framework allows for the standardization of implementation-related constructs and is able to assess multiple sides of the implementation and combines both structural and meaning elements.14 The framework was also chosen to assure continuity between the previously published article and would make for easy comparison.10 The data extracted from the included articles was put within this framework. A total of 4 domains were chosen relevant for the review question: Innovation characteristics, Individual characteristics (split up in patient/caregiver characteristics and Healthcare professional characteristics), and the inner-, and outer setting.

# Results

**A total of 1036 results were identified. Results were filtered to >2018, indicating articles published in or after 2018, and therefore not included in the previous review. A total of 349 results remained. 242 studies were excluded, leaving 107 articles that were assessed for eligibility based on the inclusion criteria.** A total of 18 articles were included in the final analysis. The flowchart is given in figure 1, a summary of study characteristics is given in table 1. Reasons for exclusion of articles were listed as well (Figure 1). A summary of article characteristics was given in **table 1,** a more extensive table can be found in **appendix 2**

Figure 1 Flow diagram of the screening process



**Table 1:** Summary of study characteristics (extensive table in appendix 1). Study types are divided into 1) remote consultations, 2) monitoring via data entered by patients, 3) passive data collection via NIV equipment.

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| Author | Type of telehealth | Study Type | study population | characteristics |
| Haulman et al. 2020.15 | Video consultations | 1 | Patients & caregivers | 38 ALS patients, 68.4% male, mean age 62 |
| De Marchi et al. 2020.12 | Video consultations | 1 | Patients | 19 ALS patients, 63% female, mean age at onset 51.48 |
| Pulley et al. 2019.16 | Video consultations | 1 | Patients | 18 ALS patients, 67% male, median age 65.5 |
| Paganoni et al. 2019.17 | Video consultations | 1 | Patients | 97 ALS patients |
| Capozzo et al. 2020.8 | Structured questionnaires via tele-cellphone or tele-video consultations | 1 | Patients & caregivers | 32 ALS patients and their caregivers, mean age 65,65 |
| Vasta et al. 2021.13 | Telephone & video consultations | 1 | Patients | 98 patients, 51% males, median age 67.6 |
| Beneteau et al. 2022.18 | Video consultations, telephone consultations, online platforms | 1 | Patients & caregivers | 6 patients (67% ALS) using AAC, 8 caregivers |
| Newton et al. 2020.19 | Video consultations | 1 | Patients | 38 ALS patients, 82% males, mean age 62.6 years |
| Rashed, 2021.7 | Telephone & video consultations | 1 | Patients | 43 ALS patients, mean age 49.4 years |
| Geronimo et al. 2019.20 | Remote PFT via video consultations | 2 | Patients & caregivers | 40 MDN patients (75% ALS) and their caregiver, 60% male, median age 61.5 |
| Hobson et al. 2019.21 | Home-monitoring via platform 'TiM' | 2 | Patients & caregivers | 40 ALS patients, 37 primary informal carers. Of patients: 70% males, mean age 60.2 years |
| Helleman et al. 2022.22 | Home monitoring of vital capacity | 2 | Patients | 33 patients (76% ALS), 79% males, mean age 60.5 years |
| Helleman et al. 2020.23 | Home-monitoring via platform 'ALS home-monitoring & coaching' | 2 | Patients | 50 patients (76% ALS), 64% males, mean age 61.4 |
| Hobson et al. 2019.24 | Home monitoring via platform 'TiM' | 2 | Patients & caregivers | 40 ALS patients, 37 primary informal carers. Of patients: 70% males, mean age 60.2 years |
| Wills et al. 2019.25 | Home monitoring via app 'NuPlanit' for food monitoring | 2 | Patients | 88 ALS patients, 53% males, mean age 56.9 years |
| Tattersall et al. 2022.11 | Remote PFT via video consultations | 2 | Patients | 25 ALS patients, 72% males, mean age 65.2 years |
| Ando et al. 2021.26 | Telemonitoring via a platform 'Careportal' | 3 | Patients | 7 patients, 71% males, mean age 63 |
| Ando et al. 2019.27 | Home monitoring via platform 'Careportal' | 3 | Patients | 13 ALS patients, 61.5% male, median age 66 years |

## Types of telehealth

The included studies reported two different types of telehealth: Distant consultations via videocall and/or telephone call, and the monitoring of patients via platforms. Platforms could include a variety of different equipment to help better assess the patient remotely.

Studies on remote consultations mostly used video consultations,12,15,16,18,19 in 3 studies telephone consultations were investigated.7,8,13 Video consultations are generally held with one healthcare professional at a time.12,18,19 One study investigated video consultations with more healthcare professionals present at the same time, if the patient preferred this.15 One study investigated the ‘store and forward’ method, in which a nurse conducted and recorded the consultation with the patient and then forwarded the relevant parts to the relevant professionals.16

Data were gathered in multiple ways while using monitoring platforms: 1) Four studies reported on monitoring platforms in which data could be entered by patients.21,23–25 2) Three studies reported on monitoring platforms where data were gathered via sensors or equipment used by the patient.11,20,22 3) Two studies reported on data being gathered passively via NIV equipment.26,27 The first four studies gathered data by letting the patient fill out questionnaires about well-being.21,23–25 These studies included one study in which nutritional counselling was performed based on nutritional data entered by the patient via a dietary app.25  The second group of studies utilized sensors and equipment to let the patient do self-assessment of certain characteristics otherwise measured in hospital, and letting them report the findings to healthcare professionals.11,20,22 Self-assessment with equipment included a number of possible tests, including forced vital capacity (FVC) measurements via a (cheap) mini-spirometer11,20,22 and weight assessment via wireless scales.21,23,24 In this type of care, specific aspects of multidisciplinary care were initiated when patient-indicated scores reached a certain threshold in disease progression.21,23,24 The last group of telehealth solutions monitors patients on the basis of passive data collection. The patient has minimal burden in this type of care. In the included studies, only remote NIV monitoring data was measured passively.10,26,27 These studies investigated data transmission from NIV equipment directly towards a digital platform on which healthcare professionals could access and assess this data to make informed clinical decisions. Direct communication between patient and healthcare professional was facilitated via this platform as well.26,27

## Barriers and facilitators for telehealth implementation in ALS

An overview of identified barriers and facilitators for successful implementation can be found in **table 2.**

### Innovation Characteristics

#### **Relative advantage**

The most reported facilitators were reduced clinical-, caregiver-, financial-, energy-, and travel burden. These facilitators were mentioned by nearly all studies, independent on the type of telehealth intervention.7,8,12,13,15,16,18,19,26,27 Apart from these finding, patients also reported feeling less abandoned during the pandemic and lockdowns associated with it,8,12 and an increased feeling of socialization and connection among patients and caregivers via digital support groups.15 One study reported that patients had the perception that teleconsultations gave them access to types of care that were at first unavailable for them.21 Three studies reported on the ability to spread out long multidisciplinary clinic days into more video consultations over multiple days.11,22,23 Healthcare professionals in one study reported that whenever something out of the ordinary was spotted during a video consultation, modification of therapy could be done with a much lower turnover time than in the classic setting of face2face care.12 The same study reported that an appointment with the proper specialist could be scheduled directly after the patient mentioned something, or the professional spotted something, facilitating offering multidisciplinary care with a lower turnover time. 12 Five studies reported that deteriorating patients were spotted earlier and received the necessary care earlier via monitoring platforms and monitoring equipment like NIV machines.11,12,20,26,27 Healthcare professionals in multiple studies mentioned both adequate and inadequate assessment of subtle patient symptoms that would determine the necessity for symptomatic care adjustments via teleconsultations.7,8,12,16,18 One study assessing NIV equipment reported that the ability to tweak NIV equipment settings in real-time and not having to wait until in-clinic visits to spot sub-optimally adjusted settings as a facilitator.27 Both NIV studies reported that (near) daily monitoring of patients made that interventions could be initiated timely by healthcare professionals and they did not have to wait for standard MDC visits. More frequent high-quality data was available to base clinical decisions on, and more continuity in care for the patient was reported.26,27

#### **Complexity and adaptability**

Generally, video consultations and monitoring measurements were not considered complex. Two studies reported that software used for video consultations was considered easy to use. 16,18 Two other studies reported that the availability of software via conventional app stores was also considered a convenience since patients could use their own devices and did not have to learn to use new equipment.23,25 Two studies reported the lack of smartphones or personal computers as a barrier to using video consultations but simple cell phones still allowed for remote consultations to continue via telephone calls.7,8 Patients reported some inconveniences with logging into the monitoring apps, partly due to the debilitating nature of ALS but generally software was considered easy to use and time burden of monitoring was low.21,23,24 The spirometers used in two studies were considered easy to handle.11,20,22

#### **Design, quality and packaging**

Two studies reported some glitches or struggles with other software programmes interfering with the software.16,18 The quality of video consultations was considered good. 8,15,16 With devices, one study reported that patients as well as healthcare professionals did experience some technical difficulties related to device malfunctioning, data transmission from the devices to central servers, and software features/bugs.26 Connectivity issues with wireless weight scales were mentioned in two studies.23,24 Having to use provided devices was sometimes considered a barrier since patients may not fully understand unfamiliar technology. One study reported that the access to telehealth services for patients using AAC devices is still under par. Software that should protect the patient by ensuring their data is secured is obstructing proper use because the software is not compatible, or at least not optimized to work with AAC systems.18 The computer software used was considered to be complex by some patients and caregivers, and easy to use by others.19 Hardware could break down because of poor store-bought quality. However, broken hardware could be reported faster to the healthcare professionals and be replaced.18

#### **Costs**

Two studies reported mentions of cost savings by patients specifically.7,8 One study calculated the costs of video consultations were twice as cheap for hospitals, and 10x as cheap for patients, but did hypothesise that with a lower threshold for patients to plan, request, or attend a consultation the frequency might increase (and therefore also the financial burden).17

### Characteristics of individuals: Patient/caregiver characteristics

#### **Self-efficacy & compliance**

One study reported patients felt comfortable discussing all problems via teleconsultations.13 Two studies, both from Mediterranean areas with lower SES reported that all patients refused video consultations, but did agree on telephone consultations.7,8 Adherence to self-monitoring was reported to be very high in all studies.22–24 Adherence fell when monitoring became too frequent. Two studies that incorporated daily monitoring reported that this was considered too often by patients.23,25 Three studies reported that patients found it confronting to see their tested values decline over time because they had more sense of what their test results meant. 11,23,25 On the other hand patients reported this as a facilitator, since they had more insights into the progression of their disease.11,21

Caregivers could often help with doing proper measurements, setting up video consultations, or filling in data.11,22,24 Two studies mentioned that patients and caregivers sometimes doubted the quality of their own measured data and felt insecure in their skills.22,24 While some studies reported that patients found apps easy to use, others reported problems with them as well and did not find them as easy to use and found learning new technological solutions to be stressful.18,24 Most of the time, over time, patients’ opinion came round after proper training with a professional or technologically literate individual.24

#### **Knowledge and beliefs**

Some studies did report that patients rather have face2face consultations or that teleconsultations were not a full replacement of face2face consultations, and considered teleconsultations helpful, but not full replacements of face2face consultations.7,8,13,19 Multiple studies reported that patients experienced an increased perceived control over- and a sense of initiative in their care. 8,11,21,23,26,27 The studies reported that patients were in closer contact with healthcare professionals via chat functions in which appointments could be requested or questions could be asked. This was confirmed as a facilitator when in another study patients explicitly mentioned the lack of feedback from a healthcare professional as a barrier.24 Adding to this was that timing for adequate support and symptom management was increased. Three studies reported that patients had the feeling that when came to the hospital, they came for a reason: Appointments were not based on arbitrary time points but based on their own disease progression.11,22,23

### Characteristics of individuals: Healthcare professionals

#### **Self-efficacy & compliance**

Monitoring Software used by healthcare professionals was reportedly easy to use apart from some glitches and modifications facilitating usage by healthcare professionals.20,24

#### **Knowledge and beliefs**

One study reported that healthcare professionals experienced fear of missing out on subtle patient clues and symptoms when only seeing them via a screen.18 Another reported barrier was the lack of an actual physical examination.16

Therapist in some studies had less confidence in the results patients communicated when using medical tools like spirometers without supervision from a professional.11,20 Professionals reported an increased sense of ability to closely monitor patients. One study reported that after data came in, disease progression or emerging symptoms were spotted quicker and modification of symptomatic therapy could immediately be done.24 Data was already available before MDC visits, increasing efficiency both during the consultation and during preparation of the consultation.23,24 Healthcare professionals in another study reported that they could not take influencing factors (e.g. patient fatigue, mood) surrounding the measurements done into account when interpreting monitoring results entered by patients remotely without supervision.22 This was not reported in the other study assessing home monitoring of vital capacity, because measurements were not done independently by the patient but in combination with a video consultation.20 The first study suggested that if measurements should be made independently, the monitoring platform should provide the option for patients to comment on influencing factors.22

### Inner setting.

Two studies reported more flexibility in scheduling of patients: Not all professionals needed to be present in house at the same time to schedule a multidisciplinary clinic day because this day could be split up into multiple consultations from home.16,18 This also led to a higher perceived efficiency in patient care. Three studies reported that more patients could be seen in the same amount of time by professionals meaning a perceived increase in scheduling flexibility.16,18,23 Two studies reported workload was similar than before telehealth.21,23 One study demonstrated that many types of appointments could take place via telehealth without affecting the quality of the consultation.19 Usually, in monitoring studies, a nurse was responsible for monitoring patient alerts. The nurse would alert the proper professional when certain thresholds in disease progression were reached, facilitating implementation because the system had to be explained to a (handful of) nurse(s) only.21–24,26 This was also seen in the store and forward method of video consultations.16 The article reporting on the store and forward method also mentioned large size videos that asked a lot of computer storage, and playback of videos leading to excessive buffering because of problems with high-speed internet within the hospital.16

### Outer setting:

#### **Patient needs and resources**

Three studies reported some troubles with internet speed and excessive buffering in consultations with patients living in more rural areas, affecting the quality of the remote consultation. 8,15,16 The availability of software via conventional app stores was also considered a convenience since patients did not have to be provided with other hardware.23,25 Patients also valued that the equipment and apps were easy to use with clear instructions and sufficient help from professionals.19,20,22–24 It was noticed in three studies that training of participants was sometimes necessary. Although this increased healthcare professional workload (barrier inner setting - available resources), this facilitated patient adherence .11,22,24 Two studies reported that when patients did not own a compatible device to install the monitoring service on, one was provided to them, facilitating implementation.20,25 The same was true in two different studies for other simple equipment.11,24

#### **External policies and incentives**

Multiple studies report that an adequate reimbursement system for telehealth is lacking. This was observed for monitoring initiatives as well as remote consultations.7,8,15–18,21,23 Two studies reported that the COVID-19 pandemic could potentially change this.7,18 One study reported on legislation issues concerning licensing of healthcare professionals practicing in different states than the patient was in. This made that professionals were sometimes not licenced in the particular state the patient was in.15 One study reported that rules and regulations including privacy legislation could actually inhibit access to telehealth for severely disabled patients. Patients experienced increased difficulty logging in to the proper systems that meet privacy standards.18

Apart from software- and hardware malfunctions, internet access as well as availability of the proper technology (smartphone, personal computer) in rural areas and areas with lower SES were reported as a barrier for the implementation of video consultations in two studies, but this could also have been a cultural phenomenon.7,8 One study mentioned the amount of different programmes, platforms, and services used by their patient group was large, and therefore healthcare professionals needed to understand many different software solutions.18

One study reported difficulty in quantifying how much monitoring initiatives actually cost per patient receiving care, because the line where the monitoring stopped and regular care began was not clear. Care associated with the monitoring initiative (messaging with the patient, phone calls) inadvertently got mixed up with regular care available to the patient making precise determination of costs for the hospital hard to assess.23 Contrary to remote consultations, there is no good cost-benefit analysis performed for monitoring platforms in ALS care, contributing to the difficulty of determining the precise cost and benefit.17

**Table 2: Overview of barriers and facilitators found in the included articles**

|  |  |  |  |
| --- | --- | --- | --- |
| Domain | Construct | B | F |
| Intervention characteristics | **Relative advantage** |  |  |
| Increase in possibilities for monitoring via technological advances |  | **F** |
| Reduced burden (clinical-, caregiver-, financial-, energy-, and travel) |  | **F** |
| Patient reported feeling less abandoned during the pandemic |  | **F** |
| Increased feeling of socialization and connection among pts and caregivers via digital support groups |  | **F** |
| Perception that patients had access to care previously unavailable to them |  | **F** |
| More flexibility in appointments. Either more HC professionals present or more spread out appointments |  | **F** |
| Earlier spotting of deteriorating patients via monitoring platforms |  | **F** |
| Adequate assessment of patient via video consult |  | **F** |
| Inadequate assessment of patient via video consult | **B** |  |
| Ability to remotely adjust patient care |  | **F** |
| More frequent accessibility to high-quality patient data |  | **F** |
| Modification of therapy could be done in a low turnover time via telehealth communication |  | **F** |
| Lower turnover time in patient symptom deterioration and appointment with a healthcare professional |  | **F** |
| More continuous care |  | **F** |
| **Complexity & Adaptability** |  |  |
| Accessible and easy to use solutions |  | **F** |
| Ability to download software on their own devices via app stores |  | **F** |
| Ability to switch to telephone consultations if no smartphone available |  | **F** |
| Inconvenience when logging into apps/using hardware due to disabling nature of ALS | **B** |  |
| Easy to handle equipment |  | **F** |
| **Design, quality and packaging** |  |  |
| Glitches or struggles with other software interfering with software used | **B** |  |
| Technical difficulties due to device malfunctioning, data transmission, and software bugs | **B** |  |
| Connectivity issues with wireless equipment (weight scales) | **B** |  |
| Under par accessibility o software when using AAC | **B** |  |
| Software was considered complex | **B** |  |
| Software was considered easy to use |  | **F** |
| Fast replacement of hardware |  | **F** |
| Both high- and low quality equipment was reported | **B** | **F** |
| **Cost** |  |  |
| Low-cost store-bought equipment usage |  | **F** |
| Reduced financial burden (missing of work of caregiver, travel costs) |  | **F** |
| 10x cheaper than in person visits (US, patient) |  | **F** |
| 2x cheaper than in person visits (US, institution) |  | **F** |
| Characteristics of individuals - patient characteristics | **Self-efficacy & compliance** |  |  |
| Patients felt comfortable discussing all problems via teleconsultations |  | **F** |
| Patients did not feel comfortable discussing problems via video consultations | **B** |  |
| Adherence to monitoring was high |  | **F** |
| Too frequent monitoring becomes burdensome | **B** |  |
| Burden of measurements increases over time due to debilitation | **B** |  |
| Patients find it confronting to see their values decline over time in monitoring | **B** |  |
| Patients gained a sense of control and disease awareness with access to their values over time |  | **F** |
| Patients doubt the results of their own measured data & their own competence | **B** |  |
| Proper training of individuals by technologically literate people increased usability for patients |  | **F** |
| **Knowledge and beliefs** |  |  |
| Some patients believe telehealth consultations are not a full replacement of face2face consultations | **B** |  |
| Patients experienced an increased perceived control over- and sense of initiative in their care |  | **F** |
| Some patients experienced lack of feedback on filled in results | **B** |  |
| Feedback on filled in results gave patients increased feeling of control over care |  | **F** |
| Patients felt that consultations were not based on arbitrary timepoints but on disease progression |  | **F** |
| Characteristics of individuals - healthcare professional characteristics | **Self-Efficacy and compliance** |  |  |
| Software was easy to use |  | **F** |
| **Knowledge and beliefs** |  |  |
| Healthcare professionals could experience FOMO during a remote consultation | **B** |  |
| No ability to take surrounding factors into account when assessing entered monitoring data | **B** |  |
| Lack of actual physical examination of patient | **B** |  |
| Less confidence in results submitted by patients | **B** |  |
| Increased perceived ability to monitor patients remotely |  | **F** |
| Quicker spotting of symptom deterioration |  | **F** |
| Amount of different apps, systems and platforms used by patients makes it hard to master them all for professionals | **B** |  |
| Increased perceived efficiency of MDC visits because of availability of data |  | **F** |
| Inner setting | **Structural characteristics/compatibility** |  |  |
| Increased flexibility in scheduling patients: not all HC professionals needed to be present at the same time |  | **F** |
| Increased workload because of training of patients | **B** |  |
| Similar workload than before telehealth | **B** | **F** |
| Many types of appointments could take place via telehealth |  | **F** |
| Some telehealth methods need high internet access and available storage in the medical center to be implemented | **B** |  |
| **Access to knowledge and support** |  |  |
| Since only the telehealth nurse needed to access the monitoring platform, training and support relatively easy |  | **F** |
| Outer setting | **Patient needs and resources** |  |  |
| troubles with internet speed and buffering | **B** |  |
| availability of software via conventional app stores |  | **F** |
| Equipment and apps were easy to use with clear instruction |  | **F** |
| Proper training of individuals |  | **F** |
| Providing/loaning of devices (tablets, spirometers, weight scales) to patients if they had none |  | **F** |
| **External policies and incentives** |  |  |
| proper reimbursement system via healthcare insurers is lacking | **B** |  |
| COVID-19 pandemic has increased usage of telehealth and therefore the need for proper reimbursement |  | **F** |
| Legislation issues between different states of patient residence and medical center | **B** |  |
| Privacy legislation is making monitoring platforms/consultation platforms less accessible | **B** |  |
| Cultural disliking towards video consultations | **B** |  |
| Difficulty in quantifying exact cost of monitoring initiatives hinders proper Reimbursement | **B** |  |

FOMO: fear of missing out

# Discussion

This systematic review summarizes the available evidence on the current use of telehealth in ALS care, and the barriers and facilitators experienced by patient, caregiver or healthcare professionals concerning telehealth implementation in ALS care. Two main types of telehealth were used in care for patients with ALS: remote consultations (either video- or telephone consultations) and monitoring (via data registered by patients, equipment, or continuous via NIV equipment). The most frequently reported facilitators were related to the relative advantage of telehealth in reducing the burden of care. This included reduction in clinical-, caregiver-, financial-, energy-, and travel burden. 7,8,12,13,15,16,18,19,26,27 Patients felt more involved in their care, more connected to their healthcare professional, and most found the equipment easy to use. Clinicians also experienced an increase in patient participation and involvement and this facilitated the access to care for patients. The platforms and software was generally easy to use for both patients and professionals. The most reported barriers were found in the outer setting, where problems with legislation and reimbursement were reported.

## Types of telehealth

While the implementation of remote consultations and -monitoring has gained attraction, no big changes in these types of telehealth have been implemented since the previous review was written.10 An increase in the availability and flexibility in teleconsultations was seen compared to the previous review. This increase might be attributed to the COVID-19 pandemic and the need to quickly implement a means of teleconsultation to not lose sight of the patient during the lockdowns. App-based self-monitoring has increased compared to the previous review, and some additions to just monitoring were added, including remote spirometer assessment and monitoring of diet.11,22,25 Passive monitoring is still only done in combination with NIV.26,27 Apart from the above mentioned innovations, no big changes in methods, measurements or data collected are seen in ALS monitoring. Comparably, in heart disease the usage of wearables has increased in care. Store bought smart watches can be used to monitor, among other biomarkers, heart rate and blood pressure.28 In diabetes, remote monitoring initiatives via wearables allowed for remote finetuning of treatment by healthcare professionals. These devices have already been implemented pre-COVID-19, but in-house appointments discussing treatment and disease progression were still standard of care.29 Results from a questionnaire in MND patients indicated that patients were very willing to have their care done more remotely, and that they would be willing to participate in clinical trials from home.30 Another survey investigating the balance between at-home and face2face consultations in different diseases indicated that 75% of patients of all disease categories that started remote consultations during COVID-19 would like to continue with them and not switch back to only in-person consultations. It is interesting to note that this decreased to 45% after one year in a follow-up study, further emphasizing that face2face consultations cannot be fully replaced in all patients.31,32 In MND care, the emergence of new biomarkers will increase the options for remote monitoring patients with more equipment. Biomarkers that could be implemented to monitor patients remotely might include activity trackers, step counters, heartrate monitors, activity trackers, and digital speech biomarkers. Nearly all these measurements can be done on smartphones. All this data is already gathered in the clinical research setting, but has not made its way into the clinic yet.33,34

## Determinants of implementation

The two most important barriers identified in this study both concerned outer setting. Problems with legislation as well as reimbursement by insurers were mentioned most often. Legislation was defined by two different aspects: 1) inaccessibility to care because of cumbersome, approved platforms that needed to be used because of privacy concerns, and 2) problems with medical licencing of healthcare professionals.15,18 Especially in the US, where medical specialists need to obtain a license per state. During the COVID-19 pandemic efforts have been made to mitigate this problem but no robust regulations are in place yet. A problem seen across multiple disciplines of care.35–37 While the usage of cumbersome software that is compliant with privacy legislation might be a barrier, patient medical data needs to be properly secured to avoid hacking and data stealing.35

Reimbursement from insurers is lacking, creating a large barrier for implementation that is observed in all different parts of the world and in different disciplines of telehealth research.7,15,23,38 One suggested solution would be the international coordination of telehealth legislation as well as reimbursement via a central (inter) national organisation coordinating both negotiations with insurers as well as with governments, patient organisations, and healthcare professionals.9

The biggest change in barriers and facilitators compared to the previous review was observed in the healthcare professionals.10 One specific barrier that was reported less than before by professionals, is the perceived lack of face2face contact. The COVID-19 pandemic probably contributed to this, since 1) there was no other option to see patients, 2) software and techniques got a big boost in availability, usability, and social acceptance, and 3) video appointments have become normalized in the professional workplace. Since patients suffering from ALS are often young and still working when they are diagnosed, video consultations might have become routine for them already. Both healthcare professionals and patients also experienced gratitude that, at least in some way, not all contact between patient and healthcare professional was lost during the various lockdowns.7,8,11,12

## Limitations

The main limitation of this study is the search strategy. Only Pubmed was consulted to search for literature while searches in other databases (e.g. Medline, Google scholar) might have identified additional publications that could be included in the systematic review. Another limitation is the types of articles included. In ALS care implementation, many initiatives are still in the pilot phase and not yet implemented in routine care (although some are). In the pilot phase some researchers and patients might be biased towards success or certain barriers might be attributed to suboptimal pilot design. Participants enrolling in pilot studies are often early adaptors with a strong affinity for innovation, that might be particularly involved in the study. Another limitation, also observed in the previous review, is the absence of primary implementation studies reporting on barriers and facilitators observed in their research. This might cause lack of reporting of barriers or facilitators in studies in which the primary focus is not on implementation. Some articles included in the search were published in late 2021/2022 but do not mention the COVID-19 pandemic as an influencing factor. Specific reported barriers and facilitators in these articles might be influenced or even directly related to the pandemic, but since this is not investigated or reported in these studies no definitive proof of the precise influencing capacity of the pandemic can be given.

## Future research

Future research should focus on creating cost-benefit analyses for monitoring platforms and -devices specifically in ALS care. Right now, some studies hypothesise that these types of monitoring will decrease the amount of necessary routine MDC visits, and even the amount of emergency hospitalizations for patients in advanced disease stages, but robust cost-benefit analyses like in video consultations, remote heart failure-, COPD, or diabetes monitoring have not been reported for ALS care yet.39–42 The difficulty reported in other disciplines with remote monitoring is that remote monitoring sometimes tends to become an addition to- rather than a replacement of regular care, increasing the cost of care per patient.43 Proper protocols should mitigate that risk. Telemonitoring did show a decrease in emergency hospitalizations in some severe diseases, which might also be true for ALS care.39,40 Since the major barriers for implementation have shifted from healthcare professionals to the inner- and outer setting, studies investigating cost-benefits could help insurers and policy makers in adjusting legislations and insurance coverage to a fitting package for patients suffering from ALS.

Another interesting area of investigation has opened up with the increase in remote and passive sensor measurements during clinical trials. With intensive monitoring of patients from home, clinical trial participation has become more accessible for disabled patients than before.44 The COVID-19 pandemic has highlighted the importance of conducting (partially) remote clinical trials, as several hundreds of trials were halted during the start of the pandemic. This has incentivised regulatory institutions to create and update guidelines for remote data collection.44,45 Another added benefit is that with high-frequency/duration measurements, clinical trial arms could be drastically decreased in size, because of higher precision-calculations of disease progression.34 Of course, the trade-off between patient burden and measurement frequency should be taken into account, but investigating at what timepoint this trade-off is optimized (highest patient adherence with most amount of measurements) could have grave implications on clinical trial duration and efficiency. With the introduction of novel remote data collection methods, including speech recordings, location data, step counters, heart rate monitors, muscle strength sensors, readily available on smartphones and wearables, an enormous abundance in data can be generated without in-clinic visits. Statistical analysis of this data is often too complex for conventional statistics. Novel AI-based tools should facilitate easier processing and analysis.45 With this new, previously unavailable data and more extensive data analysis, new biomarkers/outcome measures for disease progression should be developed. These biomarkers in disease progression might have potential to be implemented in routine care for patients as well. This might further decrease burdensome hospital visits in patients suffering from ALS. Disease progression could be monitored even more accurately from home, making even more accurate care decisions based on disease progression instead of questionnaires, routine visits, and arbitrary timepoints.

# Conclusion

This systematic review of telehealth applications and the barriers and facilitators associated with their implementation demonstrated that telehealth has further penetrated into multidisciplinary care for patients with MND. While previously patients needing ventilation benefitted most from telehealth, usage is now more widespread across all patients. This includes patients using AAC, albeit more complex in routine use for this group. While patients were already enthusiastic about telehealth, this review shows that healthcare professionals have also gained a more positive attitude towards telehealth, with fewer studies reporting barriers and more studies reporting facilitators. The main barriers for implementation are currently seen in the outer setting. Legislation is sometimes outdated, cumbersome, or necessary, hindering or slowing down implementation. Robust cost-benefit analyses for monitoring initiatives in ALS care are currently lacking, hindering, among other aspects, proper reimbursement by insurers.

Telehealth for patients with MND is focussed on remote consultations and -monitoring. Patients suffering from other chronic diseases like heart disease or diabetes have seen an increased use of low-burden passive monitoring. Similar innovations have not been widely implemented in ALS care. This might partially be due to because of lacking biomarkers for disease progression that can be linked to standard smartphones and wearables. Clinical trials have currently been started up in ALS patients using these devices, results from these might lead to new biomarkers in ALS disease progression. Via these clinical trials new possibilities for remote monitoring of MND patients will emerge. Patients as well as healthcare professionals will welcome these innovations with open arms.

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# Supplementary data

## Appendix 1: Search term on pubmed

**(((((("Amyotrophic Lateral Sclerosis"[Mesh]) OR ((Amyotrophic Lateral Sclerosis[Title/Abstract]) OR ALS[Title/Abstract])) OR "Motor Neuron Disease"[Mesh:noexp]) OR ((Motor Neuron Disease\*[Title/Abstract]) OR motor neuron disorder\*[Title/Abstract])) OR MND[Title/Abstract]))**

AND

**"Telemedicine"[Mesh] OR "Telenursing"[Mesh] OR "Cell phone"[Mesh] OR "Cell Phone Use" OR "Medical informatics"[Mesh] OR "Computers, handheld"[Mesh] OR "Mobile Applications"[Mesh] OR econsult\*[tiab] OR e-consult\*[tiab] OR ediagnos\*[tiab] OR e-diagnos\*[tiab] OR mobile health\*[tiab] OR mhealth\*[tiab] OR m-health\*[tiab] OR telehealth\*[tiab] OR tele health[tiab] OR remote consult\*[tiab] OR teleconsult\*[tiab] OR tele consult\*[tiab] OR telecounsel\*[tiab] OR tele counsel\*[tiab] OR remote counsel\*[tiab] OR distance consult\*[tiab] OR distance counsel\*[tiab] OR distant counsel\*[tiab] OR distant consult\*[tiab] OR telenursing[tiab] OR tele nursing[tiab] OR telerehabilitation[tiab] OR tele-rehabilitation[tiab] OR telediagnos\*[tiab] OR tele-diagnos\*[tiab] OR telemedic\*[tiab] OR tele medic\*[tiab] OR telemonitor\*[tiab] OR tele monitor\*[tiab] OR ehealth\*[tiab] OR e-health\*[tiab] OR telecare[tiab] OR tele care[tiab] OR digital health[tiab] OR app[tiab] OR apps[tiab] OR smartphone\*[tiab] OR phone app\*[tiab] OR telephone app\*[tiab] OR mobile app\*[tiab] OR mobile technolog\*[tiab] OR health technolog\*[tiab] OR health application\*[tiab] OR webportal\*[tiab] OR patient portal\*[tiab] OR patient platform\*[tiab] OR digital platform\*[tiab] OR digital system\*[tiab] OR e-coach\*[tiab] OR wearable\*[tiab] OR home monitor\*[tiab] OR homemonitor\*[tiab] OR self monitor\*[tiab] OR selfmonitor\*[tiab] OR tele\*[tiab]**

## Appendix 2: Study characteristics of included studies

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| Author | Publication | Type of telehealth | Study Type | study design | recruitment strategy | study population | characteristics |
| Haulman et al. 2020 | The Use of Telehealth to Enhance Care in ALS and other Neuromuscular Disorders | Video consultations | 1 | implemented as part of routine care, by option | Opportunity sampling | 61 patients & caregivers | 38 ALS patients, 68.4% male, mean age 62 |
| De Marchi et al. 2020 | Telehealth approach for amyotrophic lateral sclerosis patients: the experience during COVID-19 pandemic | Video consultations | 1 | Pilot study | Contacting all patients with MC appointments (opportunity sampling) | 19 patients | 19 ALS patients, 63% female, mean age at onset 51.48 |
| Pulley et al. 2019 | Multidisciplinary amyotrophic lateral sclerosis telemedicine care: The store and forward method | Video consultations | 1 | Pilot study | Florida-based patients suffering from MND (opportunity sampling) | 18 patients | 18 ALS patients, 67% male, median age 65.5 |
| Geronimo et al. 2019 | Evaluation of remote pulmonary function testing in motor neuron disease | Remote PFT via video consultations | 2 | Pilot study | Convenience sample | 40 patient/caregiver teams | 40 MDN patients (75% ALS), 60% male, median age 61.5 |
| Ando et al. 2021 | Experience of telehealth in people with motor neurone disease using noninvasive ventilation | Telemonitoring via a platform 'Careportal' | 3 | semi-structured interviews after a clinical trial | Opportunity sampling | 7 patients | 7 patients, 71% males, mean age 63 |
| Vasta et al. 2021 | Telemedicine for patients with amyotrophic lateral sclerosis during COVID-19 pandemic: an Italian ALS referral center experience | Telephone & video consultations | 1 | implemented as part of routine care | Opportunity sampling | 98 patients | 98 patients, 51% males, median age 67.6 |
| Beneteau et al. 2022 | Telehealth experiences of providers and patients who use augmentative and alternative communication: healthcare equality for patients using aac are lacking | Videoconsultations, telephone consultations, online platforms | 1 | Semistructured, online interviews with 6 adults using AAC and 8 clinicians who provide telehealth |  | 6 patients, 8 caregivers | 6 patients (67% ALS) using AAC, 8 caregivers |
| Hobson et al. 2019 | Process evaluation and exploration of telehealth in motor neuron disease in a UK specialist centre | Home-monitoring via platform 'TiM' | 2 | Process evaluation within a randomised, pilot and feasibility study | Opportunity sampling in patients currently receiving MND care | 40 MND patients, 37 primary informal carers | 40 ALS patients, 70% males, mean age 60.2 years |
| Helleman et al. 2022 | Home-monitoring of vital capacity in people with a motor neuron disease | Home monitoring of vital capacity | 2 | prospective cohort study | Opportunity sampling in patients currently receiving MND care | 33 patients | 33 patients (76% ALS), 79% males, mean age 60.5 years |
| Helleman et al. 2020 | Telehealth as part of specialized ALS care: feasibility and user experiences with "ALS home-monitoring and coaching" | Home-monitoring via platform 'ALS home-monitoring & coaching' | 2 | Prospective single center cohort study | Contacting all patients with MDC appointments (opportunity sampling) | 50 patients | 50 patients (76% ALS), 64% males, mean age 61.4 |
| Hobson et al. 2019 | Using telehealth in motor neuron disease to increase access to specialist multidisciplinary care: a UK-based pilot and feasibility study | Home monitoring via platform 'TiM' | 2 | single-center, mixed-methods, randomised, controlled pilot and feasibility study | Opportunity sampling in patients currently receiving MND care | 40 MND patients, 37 primary informal carers | 40 ALS patients, 70% males, mean age 60.2 years |
| Wills et al. 2019 | Nutritional counseling with or without mobile health technology: a randomized open-label standard-of-care-controlled trial in ALS | Home monitoring via app 'NuPlanit' for food monitoring | 2 | randomized open-label, standard-of-care-controlled, single-center clinical trial | Opportunity sampling in patients currently receiving MND care | 88 pts, of which 78 were included in analysis | 88 ALS patients, 53% males, mean age 56.9 years |
| Paganoni et al. 2019 | Adjusted cost analysis of video televisits for the care of people with amyotrophic lateral sclerosis | Video consultations | 1 | retrospective analysis | Opportunity sampling in patients currently receiving MND care | 97 patients | 97/97 ALS patients |
| Capozzo et al. 2020 | Telemedicine is a useful tool to deliver care to patients with Amyotrophic Lateral Sclerosis during COVID-19 pandemic: results from Southern Italy | Structured questionnaires via tele-cellphone or tele-video consultations | 1 | retrospective cohort analysis | Opportunity sampling of active patient group | 32 patients | 32 ALS patients and caregivers, mean age 65,65 |
| Tattersall et al. 2022 | The patient's perspective of remote respiratory assessments during the COVID-19 pandemic | Remote PFT via video consultations | 2 | retrospective cohort analysis | patients that neede respiratory assessment or monitoring and were open for it were included | 25 patients | 25 ALS patients, 72% males, mean age 65.2 years |
| Ando et al. 2019 | Incorporating self-reported questions for telemonitoring to optimize care of patients with MND on noninvasive ventilation (MND OptNIVent) | Home monitoring via platform 'Careportal' | 3 | Exploratory pilot study | opportunity sampling | 13 patients | 13 ALS patients, 61.5% male, median age 66 years |
| Newton et al. 2020 | Excellent reliability of the ALSFRS-R administered via videoconferencing: A study of people with motor neuron disease in Scotland | Videoconsultations | 1 | study with 2 groups, one intervention, one not | convenience sample | 38 patients | 38 ALS patients, 82% males, mean age 62.6 years |
| Rashed, 2021 | How COVID-19 pandemic changed our management strategies for amyotrophic lateral sclerosis (ALS) patients: Egyptian study | Telephone & video consultations | 1 | Prospective cohort study | all patients in cohort included | 43 patients | 43 ALS patients, mean age 49.4 years |