Incorporating self-reported questions for telemonitoring to optimise care of patients with MND on non-invasive ventilation (MND OptNIVent)

Authors

Hikari Ando: (corresponding author): Department of Respiratory Medicine, Aintree University Hospitals NHS Foundation Trust, Longmoor Lane, Liverpool, L9 7AL, UK

Tel: 0151 529 8127 Email: hikari.ando@nhs.net Helen Ashcroft-Kelso: Physiotherapist Department, Aintree University Hospital NHS Foundation Trust, Longmoor Lane, Liverpool, L9 7AL, UK

Tel: 0151 529 4015 Email: helen.ashcroft@aintree.nhs.uk

Rob Halhead: Docobo Ltd., 21 High Street, Bookham, Leatherhead, KT23 4AA, UK Tel: 01372 459 866 Email: rob.halhead@docobo.co.uk

Carolyn A Young: The Walton Centre NHS Foundation Trust, Lower Lane, Liverpool, L9 7LJ, UK Tel: 0151 529 5711Email: carolyn.young@thewaltoncentre.nhs.uk

Biswajit Chakrabarti: Chest Centre, Aintree University Hospitals NHS Foundation Trust, Longmoor Lane, Liverpool, L9 7AL, UK

Tel: 0151 529 8944 Email: biswajit.chakrabarti@aintree.nhs.uk

Peter Levene: Docobo Ltd., 21 High Street, Bookham, Leatherhead, KT23 4AA, UK Tel: 01372 459 866 Email: peter.levene@docobo.co.uk

Rosanna Cousins: Department of Psychology, Liverpool Hope University, Liverpool, L16 9JD, UK Tel: 0151 529 3178 Email: <u>cousinr@hope.ac.uk</u>

Robert M Angus: Chest Centre, Aintree University Hospitals NHS Foundation Trust, Longmoor Lane, Liverpool, L9 7AL, UK

Tel: 0151 529 3654 Email: <u>Robert.angus@aintree.nhs.uk</u>

Biographical note:

H Ando: She is a postdoctoral researcher. She has been involved in research for the last eight years, focusing on the well-being of individuals with motor neurone disease and multiple sclerosis. Her work has been shared through publications and presentations at conferences. **H Ashcroft-Kelso:** As a Clinical Specialist Home NIV Physiotherapist, she is expert in the management and establishment of patients on NIV with a variety of conditions including MND. This includes an expert understanding of analysis of patient ventilator interactions, patient and carer training and independent assessment of patients.

R Halhead: Mr Halhead has 25 years of experience in communications and systems integration. He held senior roles with BT, GE, NTL, Vodafone and two start-ups. He spent the last 8 years in telehealth, consulting with Vodafone in Mobile Health, followed by Oxford University mHealth spin-out, t+ Medical, before joining Docobo 4 years ago as Chief Operating Officer.

CA Young: Professor Young co-ordinates major clinical and research programmes in MS and MND. Nationally, she is Deputy Director of the UK MND Clinical Studies Group and on the Association of British Neurologists MS panel. Professor Young serves on a number of committees and advisory panels, including the National Institute of Health and Clinical Excellence (NICE) Appraisal Committee.

B Chakrabarti: Dr Chakrabarti is a consultant respiratory physician with an special interest in non-invasive ventilation, difficult weaning, sleep medicine, medical thoracoscopy, pleural diseases and endobronchial ultrasound. He has an extensive experience both as a clinician

and academic researcher.

P Levene: He has held senior position in the NHS in clinical measurement and has more than 40 years' experience in the medical device industry. He has led clinical research in international industrial settings, transforming outcomes into new product designs, service models and applied these through sales, marketing and strategic business development in both national and global operations. Previously he held positions on national committees tasked with the development of standards.

R Cousins: An associate professor of Health, Department of Psychology at Liverpool Hope University in Liverpool, UK. Her teaching is focused on occupational health psychology and health promotion, whilst her research focuses on promoting psychological health in neurological illnesses, recognising the influence of caregiving variables, as well as patient variables on outcomes for both.

RM Angus: Dr Angus is a consultant respiratory physician and is the lead clinician for the ventilation service at University Hospital Aintree. He has been involved with NIV since its first clinical use, introducing NIV to Aintree over the last 20 years. He has scoped the NICE guidance on NIV in MND and was part of the working group which prepared the guidelines. He has extensive experience technically in ventilation and in service development and delivery.

Abstract

Objective: Previous studies suggest a positive impact of telehealth in the care of people with motor neurone disease/amyotrophic lateral sclerosis (MND/ALS). This study reports the development of self-reported questions for telemonitoring, using a tablet-based device Careportal[®], in the care of patients with MND on non-invasive ventilation (NIV) and its initial impact.

Methods: The study consisted of a question development phase and an evaluation phase of the use of Careportal[®]. The development phase employed a modified Delphi process. The evaluation phase involved a 24-week pilot study with 13 patients (median age=66; median illness duration=14m), who were using NIV. The participants completed overnight oximetry and self-report questions via Careportal[®] each week, generating interventions where required. Patient-ventilator interaction data were monitored and the revised ALS Functional Rating Scale (ALSFRS-R) was completed.

Results: Telemonitoring encompassing the newly developed 26-item symptom questions showed good feasibility and validity. During the evaluation phase, 61 interventions were made for 10 patients, including seven patients who had routine clinic appointments during the trial to optimise care. ALSFRS-R showed significant illness deteriorations. Blood oxygen saturation (SpO₂) levels were maintained, time ventilated and inspiratory pressures increased during the trial.

Conclusion: The MND OptNIVent question set together with weekly ventilator and oximetry monitoring facilitated the maintenance of ventilation and SpO₂ levels despite illness progression. The use of the question set, and devices such as Careportal[®], facilitate care and may further enable a single point of contact for patients from which clinicians may offer proactive interventions to optimise care.

Keywords: questionnaire, telemonitoring, telehealth, NIV, respiratory symptoms

Introduction

Current UK National Institute for Health and Care Excellence (NICE) guidance recommends tailored care for people with motor neurone disease/ amyotrophic lateral sclerosis (MND/ALS)[1], with a single point of contact, flexible support, and regular monitoring.

Telemedicine may be used to assist diagnosis, symptom monitoring and management; telemonitoring allows the collection of physiological data from a patient. "Telemedicine" may be used as an umbrella term for all electronic technologies for communications and information exchange for the delivery of healthcare services [2]. However, the term is used by some to refer to services delivered by physicians only, whereas "telehealth" is used to refer to services provided by wider healthcare professionals [3]. Acknowledging the variety of definitions of telemedicine, the World Health Organization (WHO) suggested that telemedicine is broadly: "The delivery of health care services, where distance is a critical factor, by all health care professionals using information and communication technologies for the exchange of valid information for diagnosis, treatment and prevention of disease and injuries, research and evaluation, and for the continuing education of health care providers, all in the interests of advancing the health of individuals and their communities" [3].

Although limited in number, studies of telehealth for the MND population suggest it is beneficial. A retrospective study examining the efficacy of telemonitoring found equivalent quality of care between the conventional in-person consultation and video-consultation with no significant difference in survival time between the groups [4]. A single-arm study of phone call follow-up of MND patients by a nurse appeared to have enabled prompt intervention, leading Vitacca et al. to suggest the usefulness of using an interview schedule for symptom monitoring, which could effectively manage both service time and costs [5,6]. The authors suggested weekly telephone calls afforded access to healthcare services that was cost effective in managing clinical exacerbations and related respiratory problems leading to the proposal that telehealth was a valid approach [7]. However, this study had no control arm. A study of weekly remote monitoring of non-invasive ventilation (NIV) compliance suggested that telemonitoring serves to reduce hospital utilisation and lower care costs when compared with a control arm [8,9]. This study found a trend to longer survival among patients using telemonitoring when compared with controls [8], it however, did not randomise the study arms, and respiratory function between the telemonitoring group and controls was significantly different at baseline, therefore the findings should be interpreted with caution.

These studies point to benefits in utilising telehealth to care for people with MND, however randomised controlled trials with adequately powered sample sizes are needed to determine the true effect of telehealth in the care of ALS/MND patients [10]. Further optimisation of care for MND NIV users may be possible through combining remote follow up – as in the nurse-led telephone study – and telemonitoring of NIV compliance and efficacy. We envisaged that data from a self-report symptom question set together with the measurement of oximetry and patient ventilator interaction would enable clinicians to remotely monitor patients and provide interventions in a timely manner. Such an approach adds the possibility of more frequent high-quality assessment, which would not necessitate travel, resulting in less effort for the patient, and greater efficiency for the clinician. Given

the potential benefits of monitoring symptoms and NIV compliance [5,6,8,9], we investigated how combining both types of information further impacts upon the care of MND patients on NIV using a remote monitoring device, CAREPORTAL® (Figure 1). Using web-based clinical database, Careportal® offers a central contact point, as recommended by NICE, facilitating working between different multidisciplinary teams, community and hospital. The device is a tablet style computer encased in an integrated cradle to facilitate user friendliness and connection to peripherals such as a pulse oximeter; data is transferred from the patient to a N3 (National Health Service secure) cloud and a base station such as a personal computer.

[Figure 1 about here]

We designed a study that utilised a bespoke question set to assess respiratory-related symptom changes and NIV related issues, combined with oximetry and patient ventilation interaction data. The investigative study was planned with a view of establishing a telemonitoring service through a call centre operated by the local Clinical Commissioning Group (CCG). The objectives were two-fold: firstly to develop bespoke questions for Careportal[®], and then to evaluate the use of the questions as a function of telemonitoring in the care of MND NIV users. In this report, we describe the development and initial evaluation of telemonitoring using the bespoke questions in terms of work generated for the clinical team. A qualitative evaluation of the impact on patients and caregiver was undertaken and will be reported separately.

Methods

The study was conducted at Aintree University Hospital National Health Service (NHS) Foundation Trust (AUHFT) in North-West England, UK. The Ventilation Inpatient Centre at the trust care for individuals with MND, who have symptoms and or evidence of respiratory impairment. NICE guidance is followed in the management of these patients. Please refer to Supplementary file (Suppl. 1) for the description of usual care provided at the study site.

Ethical concerns

Potential participants were given a participant information sheet and written consent was obtained after participants had a chance to ask any questions. The study received favourable responses by the local Ethics Committee and by Research and Development department at the study site (14/WA/1199).

Participants

Opportunity sampling was used for recruitment. Potential participants were identified by the clinical care team from the MND NIV user database held at the Ventilation Inpatient Centre at the study site. Inclusion criteria for recruitment were: confirmed diagnosis of MND with respiratory muscle weakness; compliant with NIV (i.e. using NIV \geq 4 hours per night); capable of informed consent; anticipated survival time of \geq 6 months. See Figure 2 for the recruitment process.

[Figure 2 about here]

Study Design

The study involved two phases: the development of bespoke questions for Careportal®, and

the evaluation of the use of the questions via Careportal® in the care of MND NIV users.

Phase 1: bespoke questions development

This phase involved initial question set development and subsequent evaluation of its feasibility and validity. As illustrated in Figure 3, a modified Delphi [11] approach was undertaken. The expert panels comprised clinicians and researchers with at least 2 years' experience of supporting people with MND. The questions featured notification system to assist clinicians with symptom monitoring for timely care (see Table 1 for details). Due to the word limit, the details of this phase are provided as supplementary data (Suppl. 2).

[Figure 3 about here]

[Table 1 about here]

Phase 2: evaluation of the use of telemonitoring

The final question set (MND OptNIVent; See Supple. 3) from Phase 1 was trialled for 24 weeks to evaluate its use for telemonitoring, including face validity and feasibility of this approach.

18 patients were identified as meeting the recruitment criteria of whom 14 patients consented to enrol into the study. All participants from the development phase were invited to participate in the validation phase except one, who had died before the start of Phase 2, resulting in the total of 13 participants. During this phase, participants were prompted to answer questions and transfer nocturnal pulse oximetry readings once a week via Careportal[®]. An integrated messaging system on Careportal[®] allowed participants to contact the clinical team any time. All participants received education in terms of symptoms suggestive of chest infections and on respiratory management; they were advised to contact their general practitioner (GP) for emergencies. All were provided with rescue antibiotics.

Feasibility and validity of questions through Careportal®

Weekly follow-up was conducted via the messaging system to identify notification failures. Feasibility was tested by assessing the average completion rate; an overall response rate greater than 80% was predetermined to indicate good feasibility.

Data collection

Demographics, illness characteristics, and information on existing routine clinic appointments were collected from medical notes for each participant, and baseline pulmonary function testing was performed at the study site.

In order to evaluate how remote monitoring affects patient-ventilator interaction (PVI) maintenance, participants posted the secure digital (SD) card located in the ventilator (Respironics BiPAP Trilogy100) weekly during the study period. Participants were asked to use NIV initially at night during sleep with a minimum of 4 hours nightly. A self-report NIV usage in hours was collected.

Telemonitoring Intervention

All data were transmitted to the study site and were monitored the next working day after participants entered the data by a non-clinical researcher, who notified a respiratory physiotherapist for further assessment on the same day if any signs of worsening of the condition were acknowledged through high priority notifications. Queries raised by participants via the messaging system were also communicated with the physiotherapist. No clinical judgement was made by the non-clinical researcher. The physiotherapist was required to respond to acute respiratory events defined as: multiple new high priority notifications for one week; one high priority notification for two consecutive weeks; suboptimal nocturnal blood oxygen saturation (SpO₂) levels for two consecutive weeks. In this study, suboptimal SpO₂ was defined as: mean SpO₂ <90%, time spent under 90% SpO₂ overnight \geq 20 minutes, and/or dips in SpO₂ per hour \geq 5. The researcher and physiotherapist also had open access to both respiratory physicians involved in the study, and if they were not available, then the respiratory consultant on call would be available for advice for the ventilation patients.

Measurement

Physical functioning of participants in daily activities was measured using the revised ALS Functional Rating Scale (ALSFRS-R) [12,13] at three points: baseline, Week 12 and Week 24. Given previous concern that it is a multidimensional scale [14], sub-scores of the three domains are also reported: bulbar (Item 1-3); motor (Item 4-9); respiratory (Item 10-12). Each item is scored from 0 to 4 with higher scores indicating better function.

Data Analysis

Numbers of alerts, follow-up interventions, and any contacts initiated by patients were recorded during the study. Descriptive analyses were conducted for the number of alerts and interventions. Timing of interventions was also assessed in relation to routine clinic appointments to evaluate how Careportal[®] may contribute to optimising care.

All weekly data were analysed using data from three points: baseline, Week 12 and Week 24. Any significant findings from 12-weekly assessment were then followed by 4-weekly evaluations and weekly analyses where appropriate. Given the small sample size with great variance of the baseline clinical characteristics, assessing medians using non-parametric tests was appropriate. Differences between time points were examined using Friedman's ANOVA and Wilcoxon signed-rank tests. Bonferroni corrections were applied where appropriate. Correlations were tested using Spearman's correlation coefficient. Exact significance p-value was used where available due to the small sample size [15]. A *p*-value of .05 was considered significant.

Results

13 participants consented. Table 2 reports baseline characteristics of the participants. Table 3 shows median values for each variable at baseline, 12 and 24 weeks. [Table 2 about here]

[Table 3 about here]

Feasibility and validity of questions through Careportal®

There were no notification failures in the weekly follow-up. Ten patients had 100%

completion rate for the questions while they were in the study, the other three completed at least 80% of the required sessions.

Illness trajectory

Total ALSFRS-R scores changed significantly over the whole trial: $X^2(2) = 8.19, p = .01$. Wilcoxon tests were used to follow up this finding (see Table 4). When the three domains of ALSFRS-R were assessed separately, significant change was observed only with bulbar domain: $X^2(2) = 7.27, p < .05$. The following Wilcoxon tests showed better bulbar function at the baseline when compared with the end point, z = -2.27, r = -.53, p < .05.

[Table 4 about here]

SpO₂, NIV adherence, and patient-NIV interaction data

88.2% of the required assessments for SpO₂ were completed by participants. The range of the completion rate per participant was 66.7% to 100% (median = 91.7%). The average time of the recording time for overnight SpO₂ was 6h 40m (median = 6h 56m) with a range of 3h 36m to 8h per participant. Over the study period, no change was observed with nocturnal pulse oximetry in terms of average SpO₂ levels, time spent <90%, or rate of 4% dip.

NIV usage was assessed 12-weekly and it changed significantly over time: $X^2(2) = 10.46, p = .003$. See Table 4 for Wilcoxon tests. Both 4-weekly assessment and weekly assessment also showed a significant change over time: $X^2(6) = 28.85, p < .001$ and $X^2(23) = 44.39, p = .005$, respectively.

Availability of PVI data varied between participants with a range from 0% (n=2) to 100% (n=1). The two participants with no PVI data withdrew from NIV or died; the technical team deleted the memory before it was retrieved. The median rate for availability was 87.5 (*IQR*=14.85). The availability of the data for Week 23 and Week 24 was particularly poor; 60% and 50%, respectively. Consequently, data analysis on PVI data was assessed from Week 1 to Week 22 only. Due to the omission of Week 23 and Week 24, assessment was tested up to Week 22 and data for three points were taken from Week 1 (n=11), Week 12 (n=10) and Week 22 (n=9).

When changes between baseline, Week 12, and Week 22 were assessed, the only significant difference was found with IPAP levels: $X^2(2) = 8.86$, p = .008. Wilcoxon tests were performed and presented in Table 4.

High priority notifications

136 high priority notifications were made during the trial with a mean of 5.7 notifications per week (range from 2 to 10). See Figure 4 for the distribution of the notifications. A total number of high priority notifications triggered per participant during the trial ranged from 0 to 29 (*Median* = 8).

[Figure 4 about here]

During the trial, average SpO₂ levels for eight participants fell under 90%; nine participants spent more than 20 minutes under SpO₂ 90%; the rate of 4% dip per hour was \geq 5 for seven participants.

Interventions

In total, 61 interventions (*Median* = 5) were made during the trial for ten (77%) participants in response to telemonitoring (see Table 5 for intervention category). Amongst the ten participants, the number of interventions varied from two to nine.

[Table 5 about here]

Of these interventions, seven (i.e. 58% of provision of NIV accessories) were requested by the participants via message system or telephone (circumventing the usual email system). Six interventions resulted from suboptimal overnight oximetry readings leading to five NIV adjustments for two participants. Elective admission was made for three participants, two to optimise chest clearance and ventilation and one for optimisation of ventilation; one had been brought to clinic but due the complexity of the interventions needed admission was offered and accepted.

Interventions and healthcare utilisation

Three participants (23%) were not assessed in routine clinic during the trial yet needed intervention. Seven (54%) had routine clinic appointments during the trial; telemonitoring for six of these participants triggered 19 interventions: NIV adjustment (n=6); new home/outpatient review (n=5); providing accessories for NIV (n=4); cough insufflator adjustment (n=2); providing second ventilator for NIV (n=1); new hospital appointment (n=1).

During the study, five participants reported taking antibiotics for chest or sinus infections on 11 occasions. Three patients made unplanned GP contacts and one of them was hospitalised with pneumonia, despite being on antibiotics. This patient admission was not identified via Careportal[®] as symptoms developed between Careportal[®] entries. Another patient utilised accident and emergency (A&E) services one week after declining the new inpatient appointment offered by the physiotherapist following consecutive high priority notifications and suboptimal SpO₂ levels. This participant withdrew from the study following the admission to A&E.

Illness trajectories and telemonitoring

One of the two deceased participants made the last entry the day before their death, triggering three high priority notifications. Previously, there had been no high priority notifications triggered by this participant; at the time they were receiving antibiotics from their GP for a respiratory tract infection. Another participant who was felt to be "preterminal" triggered two high priority notifications as their last entry, leading the physiotherapist to arrange a home visit with a view to reviewing interventions; the visit was for two weeks later but the patient died before the visit.

Amongst the 10 survivors, no correlations were found between the total number of interventions and total ALSFRS-R scores or any three sub-scores of ALSFRS-R. Number of interventions was significantly negatively related with illness duration: $r_s(10) = -.74$, p = .014 and NIV duration, $r_s(10) = -.69$, p = .028.

Impact of telemonitoring on clinical team

Twelve (19.7%) of the 61 interventions were to arrange a new appointments i.e. clinical review. Three of the 12 appointments arranged were subsequently cancelled by patients; the resulting nine face-to-face contacts initiated 31.1% of all interventions made during the trial. 16.4% of interventions were phone calls to request advice from other professionals; 11.5% of interventions (i.e. seven treatment adjustments) took place during existing hospital visits since these visits fell within a reasonable timescale for response to alerts; 21.3% of interventions were equipment dispatch.

Discussion

This study reports the development of a 26-item self-report MND OptNIVent question set for weekly remote monitoring of individuals with MND on NIV. The development of the questions allowed formatting for telemonitoring. The potential benefit of regular monitoring of general MND symptoms through self-report questions has been recently suggested by Hobson *et al.* [16]. In the current study, we demonstrated a self-question set with a focus on the respiratory management to have face validity and good feasibility.

The evaluation phase showed weekly telemonitoring through the Careportal[®] to be effective in prompting changes allowing optimisation of the ventilatory support of patients with MND on NIV, in terms of SpO₂ levels and ventilation. In particular, ten of the thirteen participants seemed to benefit from close symptom monitoring and received interventions to optimise their care outside their existing hospital appointments. Only six interventions were triggered solely by suboptimal overnight oximetry readings, highlighting the importance of symptom review in addition to physiological data. This work demonstrates that the symptom related questions communicated here via Careportal[®] enabled clinicians to remotely monitor patient's symptoms effectively and provided a means to offer timely and appropriate support or intervention. Furthermore, participants utilised the message system to communicate their needs, suggesting the importance of combining all the aspects to optimise care for this patient group.

More than two-thirds of participants required interventions before and after existing routine appointments, suggesting telemonitoring has the potential to replace some traditional reviews and link the patient more effectively to their care team. The combination of education, availability of rescue antibiotics plus telemonitoring allowed effective management of intercurrent illnesses. Interestingly, one participant was admitted within days of declining the offer of an admission for adjustment of management again suggesting that the telemonitoring service was effective in identifying their deterioration to clinicians. The perceived effectiveness of care through telehealth resulting from frequent contacts with patients has been reported among patients with differing conditions [17-19]. Such continual monitoring may be particularly beneficial in optimising care in patients with MND on NIV given that there were many interventions required. Of note, during this time, there were no significant changes in the respiratory domain sub-score of ALSFRS-R, possibly reflecting a ceiling effect [14] or perhaps the effectiveness of the support. In contrast, the self-report questions were found effective in detecting weekly changes of symptoms, enabling clinicians to provide timely care.

Although 61 interventions were generated through telemonitoring, most interventions were

completed via telephone calls with only nine face-to-face reviews taking place over the trial period. It was also noted that when a home visit was undertaken in response to issues highlighted, as the clinician had access to the patient's equipment, multiple interventions could take place at one time, which may otherwise have required several outpatient attendances. While a comprehensive assessment of the impact of telehealth on health economics needs to be undertaken, these observations suggest the approach is likely to be feasible economically.

The current study found the total number of interventions to be negatively related with illness duration and NIV usage time; the longer a patient had MND or NIV, the less they needed interventions. Although this seems to contradict the general understanding of this degenerative illness, its course is known to vary [20]. In particular, the current study included three individuals who displayed slower progression (i.e. > 30 months from onset at the baseline). Only two interventions were prompted during the trial for one of these three patients, whilst the other two required no intervention. The findings suggest this patient subgroup require less input from clinicians perhaps due to their stable conditions or to well-implemented support systems over years. Telehealth such as Careportal[®] appears to benefit this patient subgroup by providing an effective communication method while also allowing monitoring against sign of deterioration for which an intervention may be needed.

It is interesting to consider that telemonitoring may particularly benefit those in the final stage of the disease when rapid deterioration may occur [21]. A telehealth service is anticipated to facilitate home-based care, which is desired by many patients towards the end of life [22], but not met in most cases in England [23]. The observation that two participants triggered multiple high priority notifications shortly before their death suggests communicating using the Careportal[®] remains viable in the terminal phase in a home environment. This type of link facilitating timely access to care should benefit not only the patient, but also their family, who often act as primary caregivers.

Limitations of the study

This question set focuses on respiratory-related symptoms as the population in question was MND patients using NIV therapy, although a question on adequacy of care, two on swallowing and the availability of the anytime message system meant that other key issues could be raised. Further question development could reflect patients' complex needs, and be tailored to patients' as suggested by Hobson *et al.* [16] to feed into MDT management for the whole ALS/MND population.

The current study involved a small sample size. While it is clear that the questions work and alerts are appropriate, further evaluation with a bigger cohort should be undertaken with the MND OptNIVent question set to determine its utility in real life, including its sensitivity and the range of multiple-choice options for answers. Ideally this would be in a randomised trial assessing telehealth against routine care.

While PVI data was available through posting, it hindered timely monitoring of NIV compliance. The loss of data particularly at Week 23 and Week 24 also confirms the importance of remote transmission of PVI data; this is now available. The benefits of such technology have been previously reported [8,9] and remote transmission of PVI is likely to

become standard.

Conclusion

We have developed a brief self-reported assessment comprised of a question set together with triggered alerts to monitor respiratory related symptoms and NIV related issues commonly experienced amongst ventilated patients with MND. Currently, 3-monthly review is recommended by NICE guidelines for people with MND. This work showed that 61.5% of patients in the study needed more frequent review, while 23.1% did not require any intervention in the 24-week period. Telemonitoring using the Careportal[®] device or a similar system integrating the MND OptNIVent question set, oximetry and PVI may allow a tailored approach to patient care, enhancing the current model of follow-up through improved patient-clinician communication.

A system like Careportal[®] could provide the single point of contact allowing signposting to the right member of the care team as communications are received. This may lead to more timely interventions where needed and reduce hospital appointments as they would not be required. It is envisaged that some routine clinic reviews could therefore be replaced by teleclinics with the data gathered from the proactive monitoring offering a comprehensive basis for a consultation. We now plan to translate this work to a routine practice setting in collaboration with one of local commissioning groups which is an early adopter of telehealth and has a call centre which can monitor the patients. This can link to the NIV and MND specialist services and also to the primary care teams via data sharing (Figure 5).

[Figure 5 about here]

We also suggest that a multicentre randomised controlled trial to evaluate the effect of telemonitoring in the care of people with MND who are on NIV should be undertaken. This should include the role of telemonitoring in acute deteriorations, a health economic evaluation with a cost optimisation assessment, comparison of key health outcomes and patient preference comparing traditional clinic reviews with proactive telehealth.

Acknowledgments

This work was collaboration between AUHFT and Docobo Ltd., who make the Careportal[®]. The work was supported by a grant from UK's Small Business Research Initiative (SBRI). The AUHFT team, in collaboration with our colleagues the Walton Centre NHS Foundation Trust developed the question set, as well as delivering the respiratory management of patients using the telemonitoring intervention.

When the question set had been iterated the display and layout of the questions was informed by Docobo's experience e.g. whether questions should be nested or single.

Docobo are pleased to have the question set published for general use.

We would like to give special thanks to our participants who graciously gave their time to take part in this study.

Declaration of Interest Statement

Rob Hallhead is a director of Docobo Ltd. who make the Careportal[®] and the software used in Smart Devices

Peter Levene is a research lead at Docobo Ltd. and helped with question structure and software development.

References

- 1 National Institute for Health and care Excellence. Motor neurone disease: assessment and management. 2016. Retrieved from https://www.nice.org.uk/guidance/ng42
- 2 Bashshur R, Shannon G, Krupinski E, Grigsby J. The taxonomy of Telehealth. Telemed J E Health 2011;17:484-494.
- 3 WHO. A health telematics policy in support of WHO's Health-For-All strategy for global health development: report of the WHO group consultation on health telematics, 11–16 December, Geneva, 1997. Geneva, World Health Organization, 1998. Cited in Telemedicine: Opportunities and developments in Member States: report on the second global survey on eHealth. (Page 9). World Health Organization 2009 ISBN: 9789241564144
- 4 Selkirk SM, Washington MO, Mcclellan F, Flynn B, Seton JM, Strozewski R. Delivering tertiary centre specialty care to ALS patients via Telehealth: a retrospective cohort analysis. Amyotroph Lateral Scler Frontotemporal Degener 2017;18:324-332.
- 5 Vitacca M, Comini L, Tentorio M, Assoni G, Trainini D, Fiorenza D, et al. A pilot trial of Telehealth-assisted, integrated care for patients with advanced amyotrophic lateral sclerosis and their caregivers. J Telemed Telecare 2010;16:83-88.
- 6 Vitacca M, Comini L, Assoni G, Fiorenza D, Gilè S, Bernocchi P, et al. Tele-assistance in patients with amyotrophic lateral sclerosis: long term activity and costs. Disabil Rehabil Assist Technol 2012;7:494-500.
- 7 Vitacca M, Paneroni M, Trainini D, Bianchi L, Assoni G, Gilè S, et al. At home and on demand mechanical cough assistance program for patients with amyotrophic lateral sclerosis. Telemed J E Health 2010;89:401-406.
- 8 Pinto A, Almeida JP, Pinto S, Pereira J, Oliveira AG, de Carvalho M. Home telemonitoring of non-invasive ventilation decreases healthcare utilisation in a prospective controlled trial of patients with amyotrophic lateral sclerosis. J Neurol Neurosurg Psychiatry 2010;81:1238-1242.
- 9 de Almeida JPL, Pinto A, Pinto S, Ohana B, de Carvalho M. Economic cost of hometelemonitoring care for BiPAP-assisted ALS individuals. Amyotroph Lateral Scler 2012;13:533-537.

- 10 Hobson EV, Baird WO, Cooper CL, Mawson S, Shaw PJ, McDermott CJ. Using technology to improve access to specialist care in amyotrophic lateral sclerosis: a systematic review. Amyotroph Lateral Scler Frontotemporal Degener 2016;17:313-324.
- 11 Jones J, Hunter D. Consensus methods for medical and health services research. BMJ 1995;311:376-380.
- 12 ALS CNTF Treatment Study Phase I-II study Group, Brooks BR, Sanjak M, Ringel S, England J, Brinkmann J, Pestronk A, Florence J, Mistumoto H, Szirony K, Wittes J, Charatan M, Stambler N, Cedarbaum JM. The ALS functional rating scale: assessment of activities of daily living in patients with amyotrophic lateral sclerosis. Arch Neurol 1996;53:141-147.
- 13 Cedarbaum JM, Stambler N, Malta E, Fuller C, Hilt D, Thurmond B, Nakanishi A. The ALSFRS-R: a revised ALS functional rating scale that incorporates assessments of respiratory function. BDNF ALS Study Group (phase III). J Neurol Sci 1999;169:13–21.
- 14 Franchignoni F, Mora G, Giordano A, Volanti P, Chiò A. Evidence of multidimensionality in the ALSFRS-R Scale: a critical appraisal on its measurement properties using Rasch analysis. J Neurol Neurosurg Psychiatry 2013;84:1340-1345.
- 15 Field A. Discovering Statistics using SPSS (3rd ed.). Thousand Oaks CA SAGE, 2009.
- 16 Hobson EV, Baird WO, Partridge R, Cooper CL, Mawson S, Quinn A, et al. The TiM system: developing a novel telehealth service to improve access to specialist care in motor neurone disease using user-centered design. Amyotroph Lateral Scler Frontotemporal Degener 2018;19:351-361.
- 17 Fairbrother P, Ure J, Hanley J, McCloughan L, Denvir M, Sheikh A, McKinstry B. Telemonitoring for chronic heart failure: the views of patients and healthcare professionals–a qualitative study. J Clin Nurs 2014;23:132-144.
- 18 Koff PB, Jones RH, Cashman JM, Voelkel NF, Vandivier RW. Proactive integrated care improves quality of life in patients with COPD. Eur Respir J 2009;33:1031-1038.
- Paré G, Moqadem K, Pineau G, St-Hilaire C. Clinical effects of home telemonitoring in the context of diabetes, asthma, heart failure and hypertension: a systematic review.
 J Med Internet Res 2010;12(2):e21.
- 20 Talbot K. Motor neuron disease: the bare essentials. Pract Neurol 2009;9:303-309.
- 21 Neudert C, Oliver D, Wasner M, Borasio GD. The course of the terminal phase in patients with amyotrophic lateral sclerosis.J Neurol 2001;248:612-616.
- 22 Baxter SK, Baird WO, Thompson S, Bianchi SM, Walters SJ, Lee E, et al. The use of non-invasive ventilation at end of life in patients with motor neurone disease: A qualitative exploration of family carer and health professional experiences. Palliat

Med 2013;27:516-523.

23 Sleeman KE, Ho YK, Verne J, Glickman M, Silber E, Gao W, et al. Place of death, and its relation with underlying cause of death, in Parkinson's disease, motor neurone disease, and multiple sclerosis: A population-based study. Palliat Med 2013;27:840-846.