"Hospital at Home" for Neuromuscular Disease Patients With Respiratory Tract Infection: A Pilot Study

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BACKGROUND: The "hospital-at-home" model may provide adequate care without an adverse effect on clinical outcome, and is generally well received by users. Our objective was to compare hospital-at-home and in-patient hospital care for neuromuscular disease (NMD) patients with respiratory tract infections. METHODS: We conducted a prospective randomized controlled trial in a university teaching hospital offering secondary care service to a population of approximately 500,000. We recruited selected NMD patients with respiratory tract infection for whom hospital admission had been recommended after medical assessment. Hospital-at-home was provided as an alternative to in-patient admission. The main outcome measures were need for hospitalization, treatment failure, time to recovery, death during the first 3 months following exacerbation, and cost of patient care. RESULTS: Among 59 consecutive NMD patients eligible for the study, 53 met the criteria for hospital-at-home. Twenty-six subjects were randomized to home care and 27 to hospital care. No significant differences were found in treatment failure (8/26 vs 13/27, $P = .19$), time to recovery (8.9 ± 4.6 vs 9 ± 8.9 d, $P = .21$), or mortality at 3 months (3/26 vs 4/27 deaths, $P = .42$) between the groups. Hospital-at-home failure was independently correlated with type of NMD ($P = .004$) with an odds ratio of failure of 17.3 (95% CI 2.1 to infinity) for subjects with amyotrophic lateral sclerosis. The total and daily direct cost of patient healthcare was significantly lower for the subjects who were successfully treated at home, compared to the hospitalized individuals. CONCLUSIONS: Hospital-at-home is an effective alternative to hospital admission for selected NMD patients with respiratory tract infections. *Key words: neuromuscular disorder; acute respiratory failure; noninvasive ventilation; hospital at home; amyotrophic lateral sclerosis; respiratory-tract infection.* [Respir Care 2013;58(12):2061–2068.

Introduction

"Hospital at home" is defined as "a service that provides active treatment by healthcare professionals, in the patient's home, of a condition that otherwise would require acute hospital in-patient care, always for a limited period."[1]

In recent years, hospital-at-home services have reached widespread diffusion for patients with a variety of conditions, including exacerbation of COPD, diabetes (uncontrolled or ketoacidosis), congestive heart failure, and

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This study was partly supported by grants from Associazione Distrofia Muscolare, Associazione Sclerosi Laterale Amiotrofica, and Unione Italiana Lotta Alla Distrofia Muscolare.

The authors have disclosed no conflicts of interest.

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DOI: 10.4187/respcare.02501

recovery from stroke or from surgery for hernia or hip replacement.[1,2]

The development of the hospital-at-home model is based on the overwhelming finding that this scheme may provide adequate care without an adverse effect on clinical outcome, and the fact that it is generally better received by users, since patients managed at home express greater satisfaction with care than those in hospital.[2,3] In addition, although the economic effects of home care programs are still controversial, studies have reported that hospital in the home can be associated with substantial cost savings.[4,5]

Respiratory tract infection is the most common cause of hospitalization for neuromuscular disease (NMD) patients, punctuating the clinical course of these individuals and triggering over 90% of episodes of acute respiratory failure.[6] Acute respiratory failure may be caused by airway mucus accumulation, atelectasis, and, ultimately, blood gas derangement, and can require intensive respiratory assistance and close monitoring.[7]

Uncontrolled clinical trials have shown that respiratory tract infections in NMD patients can be effectively managed by a home-care protocol based on the combination of continuous noninvasive ventilation (NIV), intensive manually and/or mechanically assisted cough, and arterial oxygen saturation (S_{pO_2}) monitoring, preventing or reversing oxyhemoglobin desaturation, and reducing the need for hospitalization, intubation, and tracheotomy.[8,9]

Based on these encouraging results and the observation that avoidance of hospital admission is a highly valued outcome for NMD individuals, maintaining the independence of the patient and his/her family and increasing their quality of life,[10] we were prompted to evaluate the efficacy and safety of a hospital-at-home model for the management of respiratory tract infections in NMD patients. To this end we analyzed the clinical course of 26 subjects with advanced NMD who developed severe respiratory tract infection requiring hospital admission and were treated at home, and compared the results with the outcomes of 27 patients who received conventional hospital care. In particular, we hypothesized that hospital-at-home for selected NMD patients might have an impact on clinical outcome similar to standard in-patient hospitalization, and that the application of the hospital-at-home model might reduce the direct costs of patient care.

QUICK LOOK

Current knowledge

"Hospital at home" is defined as a service that provides active treatment by health care professionals, in the patient's home, of a condition that otherwise would require hospitalization.

What this paper contributes to our knowledge

Hospital at home was an effective alternative to hospital admission for selected patients with neuromuscular disease and respiratory tract infection. The total and daily direct cost care was significantly lower for the patients who were successfully treated at home, compared to the hospitalized patients.

Methods

This study was approved by the institutional review board of our ethics committee.

We evaluated the clinical course of 26 NMD subjects suffering from severe respiratory tract infection who were treated with a hospital-at-home program, and compared their outcomes with the outcomes of a population of 27 subjects who were hospitalized. The subjects were provided with written information about the study and gave their informed consent to be assisted by the hospital-at-home program; those younger than 18 years of age reached this decision in accordance with their parents.

Subjects

We conducted the study at the Respiratory Pathophysiology Division of the City Hospital of Padova. All consecutive NMD patients who were referred to the emergency department of our hospital or to the out-patient clinic of our division between January 2009 and December 2011 with respiratory tract infection and urgent need for hospitalization were recruited.

The diagnoses of NMD were based on standard clinical, enzymatic, electromyographic, DNA, and biopsy data. The diagnosis of respiratory tract infection was based on the presence of one or more of the following symptoms or signs: fever, throat irritation or sore throat, hoarseness, and cough. The accumulation of airway mucus was defined as the coexistence of auscultatory ronchi and oxyhemoglobin desaturation ($S_{pO_2} < 95\%$).[8,11] The diagnosis of pneumonia was based on the concomitant presence of infiltrates on chest x-ray. Urgent need for hospitalization was defined as the occurrence of one of: difficulty in breathing; need for continuous noninvasive ventilatory support; or oxyhemoglobin desaturation with need for assisted cough.[8]

We excluded patients showing any of the following exclusion criteria: requirement for critical care with 24-hour surveillance; living outside the geographic area covered by our district nurse service; no non-professional caregivers or caregiver networks at home; and presence of an advance directive declining to undergo intubation and/or cardiopulmonary resuscitation.

Table 1. Anthropometric, Clinical, Pulmonary Function, and Blood Gas Data at Study Entry

	Hospital-at-Home $n = 26$	Hospitalized $n = 27$	P
Male/female, no.	17/9	24/3	.051
Age, y	44.6 ± 20.4	46.7 ± 20.2	.79
Body mass index, kg/m^2	21.4 ± 6.5	21.8 ± 4.2	.77
Type of neuromuscular disease, no.			
Amyotrophic lateral sclerosis	8	13	.29
Other	18	14	.39
Previously on long-term NIV, no.	20	24	.30
Hospitalizations in past 3 y, no.	1.04 ± 1.02	1.00 ± 1.02	.85
Cardiomyopathy and/or arrythmia, no.	7	9	.76
FVC, L	1.42 ± 1.01	1.22 ± 0.83	.48
Peak expiratory flow, L/s	2.77 ± 2.05	2.28 ± 1.36	.59
P_{aO_2}, mm Hg	69.2 ± 6.7	66.8 ± 7	.21
P_{aCO_2}, mm Hg	48.8 ± 4.5	51.8 ± 5.9	.11
S_{aO_2}, %	93.7 ± 2.4	93.3 ± 2.9	.50
Hypercapnia ($P_{aCO_2} > 45$ mm Hg), no.	19	21	.76
Pneumonia, no.	8	18	.01
Fever (temperature > 38°C), no.	14	13	.76
Leukocytosis (white blood cell count > 12,000 × 10^9/L), no.	12	12	> .99

± Values are mean ± SD.

Recruited subjects were randomized in a ratio of 1:1 to the hospital-at-home group and the hospitalized group, using blinded sealed envelopes.

Measurements

Baseline characteristics of the 2 groups are compared in Table 1. The following data were recorded at study entry: anthropometrics, type of NMD (listed as amyotrophic lateral sclerosis [ALS] or other NMD, due to peculiar clinical features of ALS), long-term use of home NIV, number of respiratory hospitalizations during the 3 years preceding recruitment, the presence of a cardiomyopathy and/or arrythmia, peak expiratory flow and FVC obtained from pulmonary function testing done within about 1 year of recruitment, arterial blood gas results, and the presence of pneumonia, fever (temperature > 38°C), or leukocytosis (white blood cell count > 12,000 × 10^6/L). All subjects were followed until recovery from exacerbation, defined as relief of respiratory distress and return of S_{pO_2} baseline to ≥ 95% during spontaneous breathing.[9] The time to recovery and the vital status at the end of the follow-up period were also recorded. For subjects in the hospitalized group, the time to recovery was considered the hospital stay. Need for hospitalization was recorded for the hospital-at-home group. Vital status at 3 months after exacerbation was also determined via telephone calls to subjects of both groups.

Intervention

Subjects in the hospital-at-home group were treated according to the following treatment protocol:

- NIV was delivered at home by a portable ventilator (Trilogy 100, Philips Respironics, Murrysville, Pennsylvania) with single-limb circuit and exhalation valve, using the continuous mandatory ventilation mode. At the start of the protocol the ventilator was adjusted to obtain a tidal volume of 10–12 mL/kg and a breathing frequency of < 25 breaths/min. The ventilator was then readjusted to maintain $S_{pO_2} \geq 95\%$. PEEP was never applied. An oronasal mask was used with all the subjects to start NIV, and then, in some cases, substituted by a nasal mask after the first few hours of NIV. Colloid dressings were placed on the major pressure points to minimize skin injury. NIV was initially delivered continuously, except for 30–60 min periods of "rest" to allow the subject to receive liquid dietary supplements, drink water, and speak. After the first 24–48 hours, if clinical conditions and blood gas exchange were satisfactory, the application of NIV was interrupted by progressively longer intervals of spontaneous breathing. In all cases, nocturnal ventilation via nasal mask was continued until the end of the follow-up period.
- Manually and/or Mechanically Assisted Cough. The following techniques were used to improve secretion clearance, depending on the subject's clinical status and

level of cooperation. Manually assisted cough was employed to provide an optimal insufflation, followed by an abdominal thrust in conjunction with the subject's coughing efforts. The portable ventilator was used to deliver the deep insufflations.[12] Mechanically assisted cough was delivered in the presence of stiffness of the chest wall (ie, severe thoracic deformity or obesity) and to subjects unable to fully perform deep insufflation. A mechanical device (Pegaso Cough, DIMA Italia, Bologna, Italy) was applied via face mask. The device has a 2-stage axial compressor that provides positive airway pressure then rapidly shifts to negative pressure, thereby generating a rapid expiration. The insufflation and exsufflation pressures and timing were independently adjusted according to efficacy and subject tolerance. Generally, pressures between 30 and −40 cm H_2O were applied.[8] Typically, a session of assisted cough was provided whenever S_{pO_2} decreased, the ventilator peak inspiratory pressure increased, or the subject had an increase in dyspnea or sense of retained secretions. Assisted cough treatments were usually repeated until one or more of the following were observed: reduction in dyspnea; reduction in breathing frequency; sputum elimination; increased S_{pO_2}. Manually assisted cough and mechanically assisted cough were administered for the first 3 days of the home care protocol by a respiratory therapist who visited the subjects each morning, and by non-professional caregivers (ie, the subject's home care attendant or a family member) trained in the application of the device. Subsequently, assisted cough was independently administered by home caregivers. The daily treatment frequency was recorded in a diary by the district nurse and/or the non-professional caregivers. At the start of the treatment protocol, the non-professional care attendants received information, instruction, and training on cough assistance from respiratory therapists of our division. The training was usually conducted over 3 consecutive days, and consisted of learning how to use NIV, mechanically assisted cough and oximetry, identification of respiratory emergencies, and training in basic life support.

- Continuous S_{pO_2} Monitoring. Hospital-at-home subjects received pulse oximeters (9500, Nonin, Plymouth, Minnesota), and their caregivers were instructed to perform oximetry feedback as needed to return S_{pO_2} to ≥ 95% by assisted coughing or NIV or both.
- Antibiotic Therapy. Standard pharmacologic treatment was used, following guidelines for the management of acute bronchitis or community-acquired pneumonia.[13,14]
- Pulmonology Visit at Home. A pulmonologist from our team visited the subjects each morning for the first 3 days, and thereafter at the discretion of the district nurses or subject's general practitioner, in order to assess the response to therapy and eventually introduce changes.
- District Nurse Visit at Home. A preexisting service of district nurses visited the subjects mornings and afternoons until recovery from exacerbation. The nurse assessed the subject's adherence and response to treatment, and could request a pulmonology visit if clinical progress was unsatisfactory. The district nurse service included 45 nurses covering an area with a population of approximately 230,000 people, and was coordinated by 2 pulmonologists.
- Other Interventions. Subject telephone access to the pulmonologists of our division was ensured. The subjects' general practitioners were faxed to inform them of the subjects being randomized to the hospital-at-home program.

The pulmonologist, nurse, respiratory therapist, or subject could request hospital admission if they felt that clinical progress was unsatisfactory. Subjects in the hospitalized group were admitted to our division and received usual care, consisting of the same drugs and all other supportive measures delivered to the hospital-at-home group, at the discretion of the ward team. Any subject who developed persistent full-time ventilator dependence and/or worsening respiratory distress and severe hypoxemia underwent tracheal intubation and, if necessary, tracheostomy.

Costs

Costs were calculated for each group from the perspective of our regional health service, such that the cost analysis was restricted to direct healthcare costs. The relevant categories to be considered in order to estimate patient costs were:

- Hospital-at-Home Group: home visit by a pulmonologist; home visit by a district nurse; home visit by a respiratory therapist; daily rental costs for mechanical cough assist and portable ventilator; antibiotic prescriptions; and telephone calls.
- Hospitalized Group: hospital stay

Data on use of categories were obtained for each subject during the follow-up period.

A second step was the evaluation of resource use. The total cost for each category was calculated as the product of the number of events multiplied by the unit cost per event. Unit costs were expressed as year 2010 prices, in Euros. Costs for pulmonology, visits by the respiratory therapist and nurse, antibiotic prescriptions, and telephone calls were directly calculated, using information on labor costs and market prices. The average hospitalization cost

Table 2. Anthropometric, Clinical, Pulmonary Function, and Blood Gas Data at Study Entry of Subjects Successfully Treated With the Hospital-at-Home Model Versus Those Who Required Hospital Admission

	Successfully Treated at Home $n = 18$	Required Hospital Admission $n = 8$	P
Male/female	13/5	4/4	.33
Age, y	35.1 ± 16.4	66.1 ± 7.7	< .001
Body mass index, kg/m^2	19.8 ± 7	24.9 ± 3.4	.07
Type of neuromuscular disease, no.			
Amyotrophic lateral sclerosis	1	7	< .001
Other	17	1	< .001
Previously on long-term NIV, no.	15	5	.33
Hospitalizations in past 3 y, no.	0.61 ± 0.7	1.63 ± 0.74	.047
Cardiomyopathy and/or arrythmia, no.	7	0	.06
FVC, L	1.3 ± 0.9	1.68 ± 1.2	.31
Peak expiratory flow, L/s	2.49 ± 1.8	3.42 ± 2.6	.44
P_{aO_2}, mm Hg	69.8 ± 6.1	68.9 ± 8.3	.38
P_{aCO_2}, mm Hg	49.5 ± 4.5	47.1 ± 4.2	.23
S_{aO_2}, %	93.9 ± 2.2	93.1 ± 3	.58
Hypercapnia (P_{aCO_2} > 45 mm Hg), no.	14	5	.37
Pneumonia, no.	3	3	.64
Fever (temperature > 38°C), no.	8	4	> .99
Leukocytosis (white blood cell count > 12,000 × 10^6/L), no.	7	5	.40

± Values are mean ± SD.

per day in the general ward and/or respiratory ICU was available in our hospital.

Study End Points and Statistical Analysis

To assess the efficacy of hospital-at-home, co-primary study end points were the need for hospitalization in the hospital-at-home group and treatment failure. Treatment failure was defined as death or the need for intubation and/or tracheostomy. Secondary end points were time to recovery, death during the first 3 months following exacerbation, and cost of care. To our knowledge, this study is the first specifically aimed at investigating the efficacy and safety of a hospital-at-home model for the management of severe respiratory tract infections in NMD patients; as a consequence, the results of any previous study could not be utilized to test an a priori hypothesis on the expected incidence or magnitude of complications associated with hospital-at-home, or to estimate an appropriate sample size.

Standard descriptive statistics were used to compare the baseline demographic and clinical characteristics. Results are expressed as mean ± SD or proportions, as appropriate. The independent unpaired Student *t* test was used to compare continuous variables with normal distribution. Nonparametric data were compared using the Mann-Whitney U test. Categorical variables were compared using the chi-square test or Fisher exact test, when required.

Variables potentially useful in predicting hospital-at-home failure were analyzed using the exact logistic regression model, considering that this procedure can adequately estimate a binary response variable with a small sample size. We calculated exact odds ratios for significant values in the univariate and multivariate models.[15] The predictor variables of interest included all data recorded at study entry. A P value of $< .05$ was considered significant.

Results

Fifty-nine consecutive NMD patients were identified as suffering from severe respiratory tract infection and therefore potentially eligible for the study. Five of those patients were excluded because they lived outside the geographic area covered by our district nurse service, and one other patient had an advance directive refusing intubation and/or cardiopulmonary resuscitation. The remaining 53 patients were recruited. Of the 53 recruited subjects, 26 were assigned to the hospital-at-home group and 27 to the hospitalized group. At baseline, the 2 groups were similar in demographic, clinical, and pulmonary function characteristics (see Table 1); however, pneumonia as a cause of acute decompensation was more frequent in the hospitalized group (8/26 vs 18/27, $P = .01$). In the hospital-at-home group, 18 (69.2%) responded well, with an uncomplicated course, and 8 required hospitalization. These 8 hospitalized individuals differed from the

Table 3. Outcomes and Direct Costs of Healthcare of Subjects Treated With the Hospital-at-Home Model Versus Hospitalized Subjects

	Hospital-at-Home	Hospitalized	*P*
Subjects who required hospitalization, no.	8		
Treatment failure, no.	8	13	.19
Time to recovery, mean ± SD d	8.9 ± 4.6	9 ± 8.9*	.21
Death at 3-month follow-up, no.	3	4	.42
Total cost of patient care, Euros	€542 ± €258.5	€8,890 ± €10,992.7	< .001
Daily cost of patient care, Euros	€65.3 ± €18.6	€1,060 ± €592.5	< .001

* For the hospitalized subjects, the time to recovery was considered the hospital stay.

others in that they were older (66.1 ± 7.7 y vs 35.1 ± 16.4 y, $P < .001$) and more frequently presented with ALS as baseline disease (7/8 vs 1/18, $P < .001$). They also had more hospitalizations during the 3 years preceding recruitment (1.63 ± 0.74/y vs 0.61 ± 0.7/y, $P = .047$) (Table 2).

With regard to reasons for hospitalization, 3 subjects in the hospital-at-home group required intubation due to persistent mucus encumbrance and severe hypoxemia, and subsequently underwent tracheostomy. Three other subjects developed persistent full-time ventilator dependence and required 24-hour surveillance in our respiratory ICU. Two additional subjects required insertion of a central venous catheter and infusion of parenteral nutrition, due to insufficient oral food intake.

The outcomes are compared in Table 3. Treatment failure, time to recovery, and mortality during the 3-month follow-up did not significantly differ (see Table 3).

By multivariate analysis, hospital-at-home failure was independently correlated with type of NMD ($P = .004$), with an odds ratio of failure of 17.3 (95% CI 2.1 to infinity) for subjects with ALS. None of the other covariates had any significant effect on hospital-at-home failure.

Total and daily direct cost of patient healthcare was significantly lower for the 18 subjects in the hospital-at-home group who were successfully treated at home than for the hospitalized subjects: €542 ± €258.5 vs €8,890 ± €10,992.7 ($P < .001$) and €65.3 ± €18.6 vs €1,060 ± €592.5 ($P < .001$), respectively. The district nurse service was the major cost for the subjects treated at home (€320 ± €118.5).

Discussion

This is the first time, to our knowledge, that the efficacy of a hospital-at-home model for managing NMD patients with respiratory tract infection has been prospectively evaluated in a controlled study. Our major conclusion is that for such patients home hospitalization is as effective as conventional hospital care, according to the observation that the proportion of individuals who had a complicated clinical course leading to intubation and/or tracheostomy was similar to that of the hospitalized group, and that both time to recovery and mortality at 3-month follow-up did not significantly differ in the 2 groups. Although the failure rate of approximately 30% for our home-based model seems high, it is in line with previous studies in the hospital setting, which reported treatment failure in the range 20–50% of subjects, and concluded that respiratory tract infections are a major cause of morbidity and mortality for this population.[16,17]

Even though our hospital-at-home scheme proved to be effective for most NMD individuals who met our inclusion criteria, the fact that more than one in 4 of our subjects could not be managed at home and needed hospital admission justifies our policy of careful home monitoring and makes it necessary to prospectively identify patients who are at risk of failing at home, in order to adopt appropriate selection criteria. Our analysis of subject characteristics associated with hospital-at-home failure concluded that the type of NMD is of critical importance, and that ALS subjects have an over 17-fold higher risk of hospital-at-home failure than do subjects with other NMDs, and their proportion among subjects who failed hospital-at-home was significantly greater than among those who responded well. This is in accord with previous reports that a noninvasive approach based on NIV and mechanically assisted cough can be problematic or even ineffective in ALS subjects, because bulbar dysfunction causes inability to protect the upper airway, which creates a risk of aspiration of food and saliva.[16,18] Our subjects who failed our hospital-at-home were significantly older than those who were successfully managed at home, although we cannot exclude that in the hospital-at-home group age may be a surrogate for other factors, such as comorbidities, which could have influenced outcome. However, concomitant chronic disease has not been found to have an important impact on the outcome of ALS patients who develop exacerbation.[19] Finally, our subjects who failed hospital-at-home had a higher hospitalization rate due to a respiratory problem during the 3 years before study enrollment, supporting the hypothesis that this "frequent exacerbator" phenotype is at high risk of failing home care and probably will require hospitalization.

Considering the changes that have occurred in healthcare in the last decade, in particular the emphasis on quality of life, cost containment, and the fact that family members have become an integral part in the home-care of severely disabled NMD patients,[20] we based our hospital-at-home model on the idea that family caregivers could acquire sufficient knowledge and skill to manage their NMD relative in case of respiratory complication. Our results show that this scheme can be effective and that non-professional caregivers can play a critical role in the transition of the care of NMD patients with pulmonary exacerbation from hospital to home, providing complex care that includes medical and nursing tasks.

In our experience, the average cost of hospital-at-home was impressively lower than hospital care: approximately €8,300 per patient, which is in line with previous studies on home treatment for COPD exacerbation and stroke.[21-23] In our study the cost savings are at least partially explained by the fact that the care was largely carried out by trained family members, reducing labor cost, which is a large proportion of hospital cost.[24]

Limitations

First, we did not collect data on patient satisfaction or the burden on caregivers; however, home has been judged to be the ideal environment for NMD individuals receiving respiratory support, since it maintains the independence of the patient and his/her family, thus increasing their quality of life.[25,26]

Second, economic evaluation was conducted from the perspective of our regional health service, so our cost analysis did not include indirect healthcare costs. This is a crucial point, since care by the family represents approximately 88% of the total cost of assisting ventilated individuals at home.[27]

Conclusions

Despite its limitations, we believe that our study provides useful information for physicians caring for NMD patients, in terms of clinical practice, which can be summarized as follows:

- Hospital-at-home, with involvement of family or other properly trained non-professional caregivers, is safe and effective for selected NMD patients with severe respiratory tract infection.
- Patients with ALS and respiratory tract infection should not be included in a hospital-at-home model, since they have a high chance of failing at home. Physicians should preferentially consider conventional hospital care for ALS patients with respiratory tract infection.
- Hospital-at-home substantially reduces costs, compared to hospitalization.

Based on these findings, we believe that effort should be expended to transform the management of respiratory tract infection in NMD individuals from hospitalization to home-care, with the caveat that proper patient selection is critical.

REFERENCES

1. Shepperd S, Iliffe S. Hospital at home versus in-patient hospital care. Cochrane Database Syst Rev 2005;(3):CD000356. DOI: 10.1002/14651858.CD000356.
2. Lemelin J, Hogg WE, Dahrouge S et al. Patient, informal caregiver and care provider acceptance of a hospital in the home program in Ontario, Canada. BMC Health Serv Res 2007;(7):130.
3. Ram FSF, Wedzicha JA, Wright JJ, Greenstone M. Hospital at home for acute exacerbations of chronic obstructive pulmonary disease. Cochrane Database Syst Rev 2003;(4):CD003573. DOI: 10.1002/14651858.CD003573.
4. Jones J, Wilson A, Parker H, Wynn A, Jagger C, Spiers N, et al. Economic evaluation of hospital at home versus hospital care: cost minimisation analysis of data from randomised controlled trial. BMJ 1999;319(7224):1547-1550.
5. MacIntyre CR, Ruth D, Ansari Z. Hospital in the home is cost saving for appropriately selected patients: a comparison with in-hospital care. Int J Qual Health Care 2002;14(4):285-293.
6. Calvert LD, McKeever TM, Kinnear WJM, Britton JR. Trends in survival from muscular dystrophy in England and Wales and impact on respiratory services. Respir Med 2006;100(6):1058-1063.
7. Vianello A, Corrado A, Arcaro G, Gallan F, Ori C, Minuzzo M, Bevilacqua M. Mechanical insufflation-exsufflation improves outcomes for neuromuscular disease patients with respiratory tract infections. Am J Phys Med Rehabil 2005;84(2):83-88.
8. Tzeng AC, Bach JR. Prevention of pulmonary morbidity for patients with neuromuscular disease. Chest 2000;118(5):1390-1396.
9. Vitacca M, Paneroni M, Trainini D, Bianchi L, Assoni G, Saleri M, et al. At home and on demand mechanical cough assistance program for patients with amyotrophic lateral sclerosis. Am J Phys Med Rehabil 2010;89(5):401-406.
10. Ambrosino N, Vianello A. Where to perform long-term mechanical ventilation. Respir Care Clin N Am 2002;8(3):463-478.
11. Bach JR. Update and perspective on noninvasive respiratory muscle aids. Part 2: the expiratory aids. Chest 1994;105(5):1538-1544.
12. Bach JR. Mechanical insufflation exsufflation: comparison of peak expiratory flows with manually assisted and unassisted coughing techniques. Chest 1993;104(5):1553-1562.
13. Wenzel RP, Fowler AA 3rd. Clinical practice. Acute bronchitis. N Engl J Med 2006;355(20):2125-2130.
14. Mandell LA, Wunderink RG, Anzueto A, Bartlett JG, Campbell GD, Dean NC, et al; Infectious Diseases Society of America; American Thoracic Society. Consensus guidelines on the management of community-acquired pneumonia in adults. Clin Infect Dis 2007;44(Suppl 2):S27-S72.
15. Hirji KF, Mehta CR, Patel NR. Computing distributions for exact logistic regression. JASA 1987;82(4):1110-1117.

16. Servera E, Sancho J, Zafra MJ, Català A, Vergara P, Marin J. Alternatives to endotracheal intubation for patients with neuromuscular diseases. Am J Phys Med Rehabil 2005;84(11):851-857.
17. Cabrera Serrano M, Rabinstein AA. Causes and outcomes of acute neuromuscular respiratory failure. Arch Neurol 2010;67(9):1089-1094.
18. Vianello A, Bevilacqua M, Arcaro G, Gallan F, Serra E. Non-invasive ventilatory approach to treatment of acute respiratory failure in neuromuscular disorders. A comparison with endotracheal intubation. Intensive Care Med 2000;26(4):384-390.
19. Bradley MD, Orrell RW, Clarke J, Davidson AC, Williams AJ, Kullmann DM, et al. Outcome of ventilatory support for acute respiratory failure in motor neurone disease. J Neurol Neurosurg Psychiatry 2002;72(6):752-756.
20. Radunović A, Mitsumoto H, Leigh PN. Clinical care of patients with amyotrophic lateral sclerosis. Lancet Neurol 2007;6(10):913-925.
21. Skwarska E, Cohen G, Skwarski KM, Lamb C, Bushell D, Parker S, MacNee W. Randomized controlled trial of supported discharge in patients with exacerbations of chronic obstructive pulmonary disease. Thorax 2000;55(11):907-912.
22. Diar Bakerly N, Davies C, Dyer M, Dhillon P. Cost analysis of an integrated care model in the management of acute exacerbations of chronic obstructive pulmonary disease. Chron Respir Dis 2009;6(4):201-208.
23. Kalra L, Evans A, Perez I, Knapp M, Donaldson N, Swift CG. Alternative strategies for stroke care: a prospective randomised controlled trial. Lancet 2000;356(9233):894-899.
24. Centers for Medicare & Medicaid Services. National health expenditure data. https://www.cms.gov/NationalHealthExpendData. *Accessed September 20, 2013. Last modified April 11, 2012.*
25. Litvak S, Heumann JE. Attending to America: personal assistance for independent living. The National Survey of Attendant Service Programs in the United States. Berkeley, CA: World Institute on Disability; 1987.
26. Goldberg AI, Frownfelter D. The ventilator-assisted individual study. Chest 1990;98(2):428-433.
27. Bach JR, Intintola P, Alba AS, Holland IE. The ventilator-assisted individual. Cost analysis of institutionalization vs rehabilitation and in-home management. Chest 1992;101(1):26-30.