Health-related quality of life in patients receiving home mechanical ventilation

Translation, adaptation and validation of The Severe Respirator Insufficiency (SRI) Questionnaire

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<th>Description</th>
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<tbody>
<tr>
<td>ALS</td>
<td>Amyotrophic lateral sclerosis</td>
</tr>
<tr>
<td>AS</td>
<td>Attendant symptoms and sleep</td>
</tr>
<tr>
<td>AX:</td>
<td>Anxiety</td>
</tr>
<tr>
<td>CRF</td>
<td>Chronic respiratory failure</td>
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<tr>
<td>BP</td>
<td>Bodily pain</td>
</tr>
<tr>
<td>COPD</td>
<td>Chronic obstructive pulmonary disease</td>
</tr>
<tr>
<td>FEV&lt;sub&gt;1&lt;/sub&gt;</td>
<td>Forced expiratory volume in one second.</td>
</tr>
<tr>
<td>FVC</td>
<td>Forced vital capacity</td>
</tr>
<tr>
<td>GH</td>
<td>General Health</td>
</tr>
<tr>
<td>HCRF</td>
<td>Hypercapnic chronic respiratory failure</td>
</tr>
<tr>
<td>HMV</td>
<td>Home mechanical ventilation</td>
</tr>
<tr>
<td>HRQoL</td>
<td>Health related quality of life</td>
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<tr>
<td>MH</td>
<td>Mental health</td>
</tr>
<tr>
<td>MHC</td>
<td>Mental health component</td>
</tr>
<tr>
<td>NIPPV</td>
<td>Non invasive positive pressure ventilation</td>
</tr>
<tr>
<td>NIV</td>
<td>Non-invasive ventilation</td>
</tr>
<tr>
<td>NMD</td>
<td>Neuromuscular disease</td>
</tr>
<tr>
<td>OHS,</td>
<td>Obesity-hypoventilation syndrome</td>
</tr>
<tr>
<td>PHC</td>
<td>Physical health component</td>
</tr>
<tr>
<td>PF</td>
<td>Physical functioning</td>
</tr>
<tr>
<td>QoL</td>
<td>Quality of life</td>
</tr>
<tr>
<td>RC</td>
<td>respiratory complaints</td>
</tr>
<tr>
<td>RE</td>
<td>Role emotional</td>
</tr>
<tr>
<td>RP</td>
<td>Role physical</td>
</tr>
<tr>
<td>SF</td>
<td>Social function</td>
</tr>
<tr>
<td>SF-36</td>
<td>Short –Form 36</td>
</tr>
<tr>
<td>SR</td>
<td>Social relationships</td>
</tr>
<tr>
<td>SRI</td>
<td>Severe Respiratory Insufficiency Questionnaire</td>
</tr>
<tr>
<td>WB</td>
<td>Psychosocial well-being</td>
</tr>
<tr>
<td>VT</td>
<td>Vitality</td>
</tr>
</tbody>
</table>
Abstract

Background: Few areas of respiratory care have advanced in the last decade as rapidly as home mechanical ventilation (HMV). In Norway the prevalence of HMV is about 20 pr. 100 000. The overriding purpose of the treatment is to maintain or increase the QoL for patients with chronic respiratory failure. For this purpose an appropriated questionnaire is needed to measure QoL in patients on HMV.

Aim of the study
- To translate and adapt the Severe Respiratory Insufficiency Questionnaire, SRI questionnaire into Norwegian,
- To test the psychometric properties of the Norwegian version of the SRI regarding reliability and validity for both patients receiving ventilation via a mask and via a tracheotomy.

Method: Accepted procedures for translation and adaptations of HRQoL instruments were followed.
SRI was psychometric tested in a HRQoL survey including patients from three counties in Norway. Both the Norwegian SRI and the SF-36 questionnaires were administered together with a specific questionnaire. In addition data was collected from the Norwegian Registry of patients receiving HMV.

Result: 127 patients responded to the questionnaire, giving a response rate of 64 %.
Reliability was obtained with Cronbach alpha coefficient >0.7 for all scales except social relationships for the non-invasive ventilated (NIV) patients, for the tracheotomised patients it was 0,52-0,89. Criterion validity obtained high correlations between SF-36v2 and SRI. Construct validity was obtained with all the hypotheses fulfilled.

Conclusion: The Norwegian version of the SRI questionnaire has good psychometric properties for non-invasive HMV. For patients receiving ventilation via tracheotomy further studies is needed to confirm the validity for this group.
1. Introduction

Few areas of respiratory care have advanced as rapidly as home mechanical ventilation (HMV) in the last decade. The total number of patients treated with HMV in Europe has increased and will continue to rise further with medical and technical advancements and as a consequence of the ageing of the population (Lloyd-Owen et al., 2005). In Norway the prevalence in HMV is about 20 pr. 100 000, which is similar to Sweden. Finland and Denmark lack national registers but is generally assumed to have somewhat lower (Finland) and higher (Denmark) prevalence’s (Fondenes, 2005).

1.1 Background

The main indication for HMV is restrictive thoracic disorders (RTD) and neuromuscular disease (NMD). The last decade the use of HMV has been extended to also include patients with hypercapnic chronic respiratory failure (CRF) caused by different underlying disorders such as progressive neuromuscular disorders, obesity hypoventilation syndrome (OHS) and chronic obstructive pulmonary disease (COPD) are (Fondenes, 2005; Lloyd-Owen et al., 2005; Mehta & Hill, 2001; Simonds, 2007).

A national centre of excellence in HMV was established in Norway 2002. A national register of patients was founded in collaboration with the Department of Thoracic Medicine, Haukeland University Hospital, to provide reliable national data on HMV, like frequency, diagnoses, symptoms, physiological findings, treatment modalities, hours of daily ventilator use, supplementary equipment, level of education, family background and the use of healthcare resources. In addition to counselling, teaching and research, the main goal of the centre is to establish and develop a skilled network of multidisciplinary health workers.

It is known that HMV impact daily living and may affect several aspects of human life (Ingadottir & Jonsdottir, 2006). Healing is not possible in the vast majority of CRF patients who receive HMV. In patients with severe disability due to chronic disease though, quality of life (QoL) may be more important than prolongation of life or improvement in physiologic outcomes (Elliott, 2002).

The overriding purpose of the treatment itself is to maintain or increase the QoL in patient receiving HMV (Fondenes, 2005). International guidelines for HMV agree unanimously on this opinion (Make et al., 1998; Robart, 1995; Simonds, 2007). The ability to evaluate health-related quality of life (HRQL) in patients with HMV is therefore of crucial importance because it is the main target and hallmark of the treatment effect.
HRQoL is considered as an important outcome parameter in patients with chronic diseases (Fayers & Machin, 2007). However, it is a challenge to measure phenomena like HRQoL in HMV patients. There are no gold standards and there is an ongoing and perpetual philosophical debate about the concept of QoL and how to define and if possible measure it. Observers frequently misjudge both symptoms and general QoL issues according to several studies. Health care personal and observers in general tend to base their judgement of overall QoL upon physical signs and symptoms (Fayers & Machin, 2007). In many disease areas, conventional clinical outcome parameters are poorly correlated with patient assessment of QoL Furthermore, studies have shown that healthcare professionals in some cases do not inform about or recommend HMV because they believe the adverse effects of the treatment to be worse than the benefit (Bach, 1995; Bach, 2002, 2004).

Traditionally, medical decisions have tended to concentrate upon mortality and morbidity, i.e. symptom relief and cure as primary outcome measures. Studies using QoL instruments may reveal other issues that are equally or more important to patients (Fayers & Machin, 2007a). The increased emphasis on QoL reflects an important shift in medical thinking. More importantly, where there is conflict between QoL versus mortality/ morbidity outcome measures, it is no longer accepted that the former should play a less important role. An implication of this being that that there may be circumstances when maintaining the patients QoL takes priority over prolongation of life or reducing morbidity. Balancing the relative importance of mortality, morbidity and QoL in medical decision making is always subjective and different conclusions may be drawn about patients who have identical medical conditions (Ahmedzai & Muers, 2005).

Several national clinical guidelines emphasize the importance of measuring QoL. Research in QoL is necessary to evaluate the quality of the health care services, evaluate outcomes and resource expenditure (Staniszeweska, 1998; Wahl & Hanestad, 2004). Measuring QoL is also important in medical decision making. QoL can be a predictor of treatment success (Fayers & Machin, 2007). Several studies have also found that QoL in combination with other factors, like pain and physical well-being, are of prognostic importance (Fayers & Machin, 2007). HMV is applied in mainly two different modes. In Norway nine out of ten patients receive treatment in a non-invasive ventilatory mode (NIV) via a nasal or facial mask, and only one out of ten patients are ventilated invasively via a tracheotomy tube with an artificial opening on the neck (Fondenes, 2005). The trend internationally is towards an increasing number of patients ventilated in non-invasive mode.
The patient group is relatively small and heterogeneous in terms of diagnosis, disease trajectory and prognosis. Consequently it is likely that multi-centre studies will be necessary to answer important questions regarding the benefit of treatment for different patient groups. A cross cultural applicable instrument is necessary for this purpose.

In the past few decades, several generic and disease-specific questionnaires have been developed for QoL assessment (Bowling, 2001; Fayers & Machin, 2007a). Many of them have been translated into Norwegian. Most of the questionnaires used in earlier studies of HMV patients were not disease or condition specific. Many aspects and conditions that are important for patients receiving HMV were not included in these questionnaires and consequently incomplete assessments may have been made (Wijkstra, Lacasse, Guyatt, & Goldstein, 2002; Windisch, Budweiser, Heinemann, Pfeifer, & Rzehak, 2008; Windisch et al., 2003a). Until very recently there were no specific questionnaires developed for the HMV group of patients. The Severe Respiratory Insufficiency (SRI) questionnaire was first presented 2003 as a multidimensional HRQL tool with high psychometric properties, specific for measuring HRQoL in patients receiving HMV. The SRI questionnaire was specific developed for patients receiving HMV in NIV mode (Windisch et al., 2003a). However, the author Windisch, and Lopez-Campos, the researcher who translated SRI into Spanish, suggested that it would be of interest to study HRQL in the tracheotomised patient, to establish whether the SRI could discriminate between the tracheotomised patient and the patients ventilated via a mask (Lopez-Campos et al., 2008).
2. Aims of the study

The aims of the study is:
- To translate and trans-cultural adapt the Severe Respiratory Insufficiency Questionnaire, SRI questionnaire into Norwegian,
- To test the psychometrics property of the Norwegian version of the Severe Respiratory Insufficiency Questionnaire, SRI regarding reliability and validity for both patients receiving ventilation via a mask and for patients receiving ventilation via a tracheotomy.

3. Home mechanical ventilation

3.1 History

One of the earliest successful attempt to ventilate a human being by a mechanical device was the use of negative pressure ventilation in 1832. It was a simple precursor to the tank ventilator or so called iron lung.

The extensive outbreaks of poliomyelitis in Europe in the beginning of 1950, with a fatal course due to respiratory muscle paralysis created a need for long-term ventilatory support. Many patients also suffered from bulbar paralysis, and in 1943 tracheotomy were proved to be a life-saving procedure for these patients. During the polio epidemic in Denmark in 1952 manual intermittent positive pressure ventilation via tracheotomy was introduced, and the results of this treatment provided the impetus for a revolution in the medical care of patients with respiratory failure. Until the late 20th century, the only techniques suitable for long-term respiratory support in the home were tracheotomy ventilation (Simonds, 2001). With the advent of bi-level positive pressure ventilators for nasal mask ventilation in the mid-eighties, an extraordinary expansion of the treatment possibilities has taken place. Largely due to the ease of use and benefits both in terms of patient comfort and cost of treatment, ventilators for non-invasive use are the mainstay of HMV today.
3.2 Epidemiology

3.2.1 Norway

Patients with poliomyelitis are still an important, but proportionally decreasing, group of patients in the Norwegian population of HMV users. Patients with NMD constitute about four out of ten patients in Norway. The largest increase in prevalence of treatment has been registered in the so-called lifestyle diseases such as Obesity-hypoventilation syndrome (OHS), which constitute one out of five patients, and COPD accounting for nearly one out of five patients. However, the diversity of diseases which are considered relevant indications is great and more than 24 different diagnostic labels have been recorded in the Norwegian registry (Fondenes, 2005).

3.2.2 Europe

A survey of the pattern of HMV users in Europe has shown large differences across countries. Overall, the HMV users were divided into one third lung and airway diseases (e.g. COPD), one third thoracic cage problems (including OHS) and one third NMD. However, there were marked differences in these proportions comparing northern vs. southern Europe. In northern Europe, the prevalence of NMD patients on HMV is much higher than those with lung disease. Southern countries had a higher proportion of users with lung conditions, mainly COPD patients (Lloyd-Owen, 2005).

3. 3 Pathogenesis

Hypoventilation implies insufficient alveolar ventilation in relation to the body’s metabolic need. It is characterized by increasing arterial carbon dioxide tension and decreasing arterial oxygen value (Jett, Spiro, & Albert, 2008). This form of CRF is mainly caused by impaired function of the “bellows” capacity of the inspiratoriske muscles to sustain an adequate ventilation in the chest wall and neuromuscular diseases. In opposition, more common types of CRF may be caused by a primary lung disease where imbalance between ventilation and perfusion of different parts of the lungs will result in a poor gas exchange. Depending on the disease and course, of development of CRF, the symptoms usually takes place gradually, at first only present during sleep and as the disease progresses, also during daytime. Conditions and diseases in which benefit from HMV are listed in Box 1.
### Box 1, Indications for domiciliary ventilation (Simonds, 2007).

<table>
<thead>
<tr>
<th>Condition</th>
<th>Level of recommendation</th>
<th>Comment</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>a) Chest wall disorders</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Scoliosis</td>
<td>C</td>
<td>RCT Unlikely to be conducted as outcome without ventilator support is fatal.</td>
</tr>
<tr>
<td>Thorakoplastikk/previous</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>tuberculosis</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Fibro thorax</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Morbid obesity</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>b) Neuromuscular disease</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Congenital</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Myopathies</td>
<td>C</td>
<td>RCT unlikely to be conducted for same reasons as stated above</td>
</tr>
<tr>
<td>Duchenne muscular dystrophy</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Other muscular dystrophia</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Spinal muscular atrophy</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Hereditary sensory neuropathies e.g.</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Charcot-Marie-Tooth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Acquired neuromuscular disease</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Old poliomyelitis</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Polymyositis</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>ALS/ motor neuron disease</td>
<td>A</td>
<td>One RTC</td>
</tr>
<tr>
<td>Cervical spinal cord lesion</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td><strong>c) Neurological disorders</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Congenital central hypoventilation syndrome</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Brainstem CVA</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>d) Obstructive lung disease</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>COPD</td>
<td>B</td>
<td>COPD: Mixed results cross over trials. RCT vs. LTOT required</td>
</tr>
<tr>
<td>Idiopathic bronchiectasis</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>Cystic fibrosis</td>
<td>C</td>
<td></td>
</tr>
</tbody>
</table>

**Comments related to evidence:** A-Supported by at least one randomized trial (RCT), B-Supported by well conducted clinical studies, C-Consensus evidence

**Abbreviations:** CVA, cerebrovascular accident, LTOT, long-term oxygen therapy

RCT = Randomized controlled trial

Because of the limitations of this study only some of the diseases listed in box 1. are described subsequently.
a) Chest wall disorders

From a pathophysiological viewpoint the main group of disorders that may induce hypoventilation is characterised as restrictive chest diseases. The term “restrictive” refers to the small volumes of air the patient is able to inhale as measured by spirometry. The reason for this might either be increased stiffness of the chest wall, secondary to or muscular weakness the latter usually due to a neurological disorder that might cause problems expanding the chest. In a 20 years follow-up study of lung function in adults with idiopathic scoliosis CRF occurred in 25% of the patients. Death was due to cor pulmonale (chronic strain of the heart due to hypoxia and increased resistance in the pulmonary vascular bed) in 30% of the patients with untreated idiopathic scoliosis. RCT is unlikely to be conducted in this group because the outcome without ventilation support in this group is fatal (Make et al., 1998; Simonds, 2007).

Obesity Hypoventilation syndrome (OHS) is defined by the development of diurnal hypercapnia in obese individuals (BMI >30 kg/m²) in the absence of other reasons for hypoventilation such as coexistent lung or neuromuscular disease. OHS is associated with a significantly greater degree of morbidity and mortality than obstructive sleep apnoea OSA primarily related to compromised respiratory and cardiac function (Berg, Delaive, Manfreda, Walld, & Kryger, 2001; Budweiser et al., 2007; Hida et al., 2003). OHS is rapidly becoming the major indication for HMV in many centres in the western world (Laub & Midgren, 2007).

b) Neuromuscular disease, (NMD)

Congenital NMD:

Duchenne muscular dystrophy (DMD) is one of the congenital NMD. DMD are genetic disorders characterized by progressive muscle wasting and weakness. As muscles degenerate over time, the person’s muscle strength declines. From teenage about 90% of the patients need assisted ventilation. This group contain 6, 4 % of the patients in the Norwegian registry (Foondenes, 2005). Without HMV, morbidity and mortality are increased highly likely towards the end of the second decade of life in patients with this disease. Nocturnal NIPPV reverses symptoms associated with poor sleep quality (Ward, Chatwin, Heather, & Simonds, 2005) and full time HMV reverses symptoms associated with daytime dyspnoea (Toussaint, 2006).
Aquired NMD:

Amyotrophic lateral sclerosis (ALS) is an acquired NMD. The annual incidence of ALS in Norway is 2 per 100,000 persons. The total number of ALS patients in Norway treated with HMV is around 60. The cause of neuronal degeneration in ALS is unknown. Most patients die either of pulmonal complications or respiratory failure {Fondenes, 2005 #151} Bulbar problems affect up to 30% of individuals at the onset of the disease and is recognized by excessive oral secretions, aspiration and choking episodes. Ventilation via a tracheotomy is the only ventilation option in patients with severe bulbar disease (Simonds, 2007).

High spinal cord injury is often acquired by a trauma. High injury, above C4, affects the phrenic nerve and thus involves the diaphragm function. The need for HMV depends on the level of the spinal cord injury. These patients constitute only about 3% of the patients in the Norwegian registry (Fondenes, 2005).

c) Other neurological disorders

Patients that may be candidates for long-term ventilation are those with absent or severely impaired spontaneous breathing efforts. These patients include those with central hypoventilation secondary to inadequate central respiratory drive (i.e., intracranial haemorrhage, cerebrovascular accidents, and central alveolar hypoventilation) (Simonds, 2007).

d) Obstructive lung disease

In patients with respiratory failure primarily due to obstructive lung disease the indication for HMV may not be as obvious as in NMD patients (Simonds, 2007).

An estimate suggests 250,000 patients with COPD in Norway. The most important risk factors are age and smoking habits (Johannessen, Omenaas, Bakke, & Gulsvik, 2005). A small but significant number of patients develop advanced disease with CRF. The evidence regarding benefit of NIV in this sub group is contradictory. The only therapy which has been shown to improve survival in patients with CRF due to COPD is long-term oxygen therapy LTOT. A Cochrane meta-analysis concluded that HMV had no consistent clinically or statistically significant effect on physiologic outcomes (Wijkstra, 2003).
There are some studies that support the use of NIV in selected patients, i.e. patients with recurrent hospital admissions with acute on CRF. (Simonds, 2007). During the last years studies have been published that find improvement in both general and condition-specific aspects of HRQL following HMV in COPD patients (Windisch, Budweiser, Heinemann, Pfeifer, & Rzehak, 2008). The issue concerning possible benefit of long-term NIV in COPD patients with CRF is important because of the high prevalence of COPD worldwide (Fondenes, 2005; Simonds, 2007).

### 3.4 Symptoms of hypoventilation

Symptoms of hypoventilation may present a wide array of physical, psychological and neurocognitive categories. Such as dyspnea, hyper somnolence, fatigue, morning- or continual headaches, sleep disturbances, awakenings with sensation of dyspnea and tachycardia, anxiety, nocturi, difficulties in concentrating, irritability, impaired intellectual function, memory impairment, muscular pain, depression, frequent nightmare, decreased libido. HMV can provide adequate ventilation and reverse these symptoms (Bach & Alba, 1990; Fondenes, 2005; Hill, Eveloff, Carlisle, & Goff, 1992; Schiavina & Fabiani, 1993).

### 3.5 Treatment

The term “ventilator” describes the main function of the equipment, which is to offer artificial ventilation to patients with impaired ability to exchange blood gases (Dybwik, 2000). Ventilators for home use have developed via two main evolutionary pathways. Originally, positive pressure home ventilators were derived from ICU types of ventilators, with similar kind of functionality and appearance. The alternative pathway of development was primarily based on equipment intended to treat patients with obstructive sleep apnoea, hypopnoea syndrome. The latter machines were of simple design and did not include a great range of modifiable parameters or alarms (Simonds, 2007). These ventilators are easily triggered by the patients own effort to inspirate and commonly used as support to the patients ventilation. The ability to fully control and compensate a complete loss of the patient’s ventilation capability however is smaller than the “ICU ventilators”. Ventilatory support increases gas transport to and from the alveoli and relives the work of breathing helping the patient using less muscles effort in the work of breath. During expiration the ventilators airflow generates a pressure that maintains potency of the upper airways and facilitates removal of mucus and secretions (Fondenes, 2005).
3.5.2 Elective indications

The appropriate timing for HMV is crucial. The elective introduction of treatment presupposes that high risk patients can be identified, that the natural history of underlying respiratory disorder is known, and that the overall impact of treatment is beneficial. Also it is essential that all non-ventilatory therapeutic options have been fully explored before embarking on respiratory support (Simonds, 2007).

3.5.3 Acute indications

Advances in medical care and the acute application of invasive mechanical ventilation have resulted in increased survival of critically ill patients, some of whom may become dependent on long-term mechanical ventilation (Make et al., 1998). On occasion acute respiratory failure may represent the initial manifestation of progressive neuromuscular disease, requiring long term hospital admission, often in an ICU.

3.5.4 Mode of ventilation

HMV can be applied in invasive and non-invasive mode (NIV). Invasive means via a tracheotomy. NIV can be applied in positive pressure ventilation (PPV) and negative pressure ventilation (which implies f.eks. pneumobelt but only a couple of patients in Norway are ventilated in this mode). The various methods of applying non invasive positive pressure ventilation, NIPPV, ventilation are -nasal mask, -facemask, -nasal plugs, -helmet-mouthpiece. Growing experience with these has proven these options to be feasible in circumstances where previously invasive ventilation was thought mandatory. This is due mainly to the simplicity and cost of the treatment mainly. NIPPV is easy to administer, it preserves a normal function of the upper airways, which allows the patient to speak, eat and swallow with less difficulty than a tracheotomy (Fondenes, 2005; Simonds, 2007).
3.6 Organizing treatment and follow-up of patients

Organizing and planning treatment and care of the HMV patient in Norway is regulated by legislation and partly addressed in several national recommendations (HOD, 1984, 2001a, 2001b, 2003, 2004, 2005). The HMV patient group is heterogeneous in many aspects, both concerning level of ventilator dependency, functional disability and the need for care given from nurses or unskilled personnel (Fondenes, 2005). Many HMV users do not require any care giving support at all and are self-sufficient as long as they use their ventilator. These patients may be working ordinary jobs or studying.

At the other end of the scale, patients who require continuous HMV usually depend on a team of caregiver personnel. Technical resource needs may also be extensive in terms of respiratory equipment such as backup ventilators, monitors, mechanical cough assist and equipment for humidification and secretion suction. The risk of fatal complications may be increased with unskilled or uneducated health-caregivers and technical malfunction due to lack of follow-up (Chatila, Kreimer, & Criner, 2001; Criner, Tzouanakis, & Kreimer, 1994). Majority of treatment accidents reported are caused by improper use of equipment due to lack of education and training of patient or caregivers (Srinivasan et al., 1998). In Norway it is estimated that about every fifth HMV user have a particular high requirement for support and care giving (Fondenes, 2005).

3.7 Ethical aspects

Medical and technical development provides us with new treatment options for patients in need of HMV. However, ethical challenges in several areas may arise as a result of these possibilities as well, both in terms of nursing-practice and in the process of deciding treatment options. The Norwegian nursing society’s professional ethical guidelines states that nursing is performed in vulnerable arenas (NSF, 2007).

Since the 1980ies principia based ethics has been the most common method for ethical interpretation in biomedical context. The four basic principles encompass:

1. The principle of autonomy or self-determination implies respecting the choices and wishes of persons who have the capacity to decide and protecting those who lack this capacity,
2. The “do not harm” principle implies that one should never harm or injure a person.
3. Beneficence means acting in the best interest of the person or doing the best to protect him/her. Promoting beneficial factors and removing harmful factors in a patient’s situation.
4. The justice-principle represents the idea of justice in politics and partings of preferred factors (Beauchamp & Childress, 2008).

In essence, providing persons with that to which they are entitled and treating similar cases similarly. Informing the HMV patient about the treatment possibilities and implications in terms of physical, emotional and social consequences invariably needs to balance the above mentioned principles. Fulfilment in practice is a process that requires experience, compassion and wisdom of the physicians, nurses and other healthcare workers involved. It is also important to be aware of the fact that different patients have difference preferences as to how much they want to know. Practical issues like living at home or in a service apartment or institution may impose ethical problems both in terms of the ability of the patient to choose and what may be the best interest of the patient. Similar questions may arise regarding the different ways to organize care giving, using personal assistants (patient has the responsibility to organize the caregivers) or home nursing (primary health care service).

HMV also may have a huge impact on the patient’s family often relatives and friends. It is important that they are properly informed as well. Furthermore, help and opportunity to express themselves about their wishes for the patient and what they might need to make the treatment successful should be provided. The documentary “Eit meiningsfylt liv i ein lam kropp/ A meaningful life in a paralyzed body (Ese et al., 2005) shows in a comprehensive manner both the challenges and solutions to some of these questions with advanced stage of ALS, and reflects the positive attitude of the patient and his caregivers. On the other hand there are studies that indicate a very high burden of care for caregivers and relatives (Kaub-Wittemer, 2003). HMV in a rapidly progressive disease like ALS implies several severe ethical challenges. As the patient gradually becomes more paralyzed in the end this could prevent him or her from any kind of communication although the intellect might be preserved. This condition is sometimes referred to as the “locked-in” state. Ethical problems regarding ventilator treatment or discontinuation of such under these circumstances have been discussed. Is it possible for the patient to decide to withdraw treatment even though the consequence of this is fatal? Would such an action be a form of euthanasia? The Norwegian ethical council of medicine has expressed written opinions regarding this situation: In general “close to the end or at the end of life the doctor must respect the patient’s right to decide… …to end or not to commence pointless treatment is not to be judged as active assistance of death or euthanasia.” (§ 5 in chapter 1 in ethical rules.).

Before ventilator treatment is commenced in patients where a rapid and severe decline of function is expected these issues should be discussed and the patient’s opinions should be
documented. In some cases, although legal status of such documents are uncertain, advance directives may be formulated. In any case, the issues need to be reassessed on a regular basis as the patients opinions may change. Also it should be clarified in advance how the patient is going to express his/her will to withdraw from active treatment. When a patient expresses this wish, there must also be a certain period given for the patient to reconsider before the final resolute is made (Førde, 2006)
Respecting patient autonomy and ensuring adequate information to make him/ her qualified to make difficult decision is an important ethical challenge. Health personnel’s own perception of what QoL is and what should be deemed a worthy life could influence decisions either by misinformation or poor advice (Bach, 2004; Fayers & Machin, 2007). This demands reflection and self-awareness about ethical choices that has direct consequences for vulnerable patient groups. These challenges could be summarized by quoting Løgstrup’s statement “As individuals, dealing with our fellow man, we a carry a piece of their destiny in our hands (Løgstrup, 1991).

3.8 National advices concerning HMV

"The right to health treatment is only relevant when the patient can expect significant gain from the treatment and when the costs are in reasonable relation to the effect of the treatment” (HOD, 2001a). The Norwegian national council for quality and prioritising in the health services this may (2008) stated evaluation and recommendations and made also several stipulations when considering HMV. Among other considerations concerning the HMV group, they requested outcome data, of effect of HMV for other groups than those with NMD and RTD. They have commented that the lack of evidence for beneficial effects in COPD and consequently recommended a strict policy against widespread use of the treatment in these patients. The council also emphasized the need to clarify the financial burden in particular regarding patients in need of 24 h caregiving. The question being to what extend society is able and willing to support homecare over longer periods of time under such circumstances (Nasjonaltråd & helsetjenesten, 2008).
4. Quality of life

4.1 History
The term QoL was first mentioned in 1920, then later on in a book about economy and welfare (Pigou, 1920). In the sixties and the seventies the development of the welfare society brought with it the need to be able to measure other qualities than just material satisfaction. The first publication of a comparison between medical treatment and the QoL was “Medicine and quality of life” (Elkington, 1966).

In nursing science the term of QoL life was established in” The cumulative Index for nursing and Allied Health Literature” from 1983 (Padilla, Grant, & Ferrell, 1992). Since 1980 there has been a rapid increase of publications related to QoL research. In Norway a several studies on QoL within a wide area of patient groups have been conducted. This indicates a growing interest for QoL related to clinical practice. Especial interest in this field of research is found at the Section of Nursing Science, Department of Public Health and Primary Health Care, University of Bergen. These studies and others are providing increasing and more systematic knowledge on how illness and reduced health affects a person’s QoL, thus giving us the opportunity to make more adapted and appropriate efforts to help.

4.2 Quality of life concept
Quality of life QoL is a broad concept. Efforts to formulate a global consensus definition have been made for several years and there is still no agreement upon a common definition. Lack of agreement in the meaning of the term might be explained by the complexity and the dynamic character of the concept and might also be due to the fact that several disciplines with different perspectives and concerns, including, sociology, philosophy, medicine and nursing are involved (Fayers & Machin, 2007). The different definitions of the concept often reflect the different researcher’s area of interest, in which the term was developed and put to practise (Wahl & Hanestad, 2004).

Many authors have focused on what is the important part of QoL. Siri Næss presents an emphasis on the subjective experience of the good and satisfactory character of life; “The QoL is to have good feelings and positive evaluations about ones own life. Good feelings can be of several kinds; joy, energy and love i.e. Positive evaluations can be understood as
being satisfied, to have self-respect and the sense of purpose to your existence “(Næss, Moum, Mastekaasa, & Sørensen, 2001).

The definition stated by Schipper and colleagues is simple and perhaps somewhat limited. “The functional effects of a disease and its following therapeutic effect on a patient as it is experienced by the patient” (Schipper, 1990).

Upon examination of theoretical articles published in the QoL field, the first theme to emerge was the multidimensional and dynamic nature of QoL. Authors also stressed that QoL is based on values and that it is dynamic and changing, depending on the context in which it is measured.

The second theme was that QoL includes an assessment of some kind. Within this theme, authors included words such as response, appraisal, measure, experience, and assessment in defining and using QoL.

The third theme to emerge centered on the subjective nature of QoL. Grouped within this theme were words like perception, well-being and satisfaction.

The final theme that developed recognized the objective nature of QoL, with authors using behaviour, functioning, and environment as measures of QoL (Haas, 1999).

To be able to use QoL terms in clinical practice it is necessary to make the concept operational in a way that makes it possible to measure (Wahl & Hanestad, 2004).

4.3 Different levels of QoL

QoL in the terms of health profession can be measured on different levels, the general, the global and the disease-specific level (Wahl & Hanestad, 2004). These three levels show the complexity of the term QoL, also how closely the QoL concept is on the general well-being of the individuals and the relation to specific symptoms caused by disease. There are different QoL definitions. Some definitions include health and the patient perspective, while others have a more global character. Term wise clarity in what is measured is necessary to be able to make a clinically valid study and to make the research relevant to treatment and care of patients.

Health professionals can have different perspectives in patient care. In medicine the symptoms and treatment of the disease itself is the primary focus. In nursing one have a tendency to be more focused on what recourses and limitations the patient meets in everyday life because of his or her disease. A this point of intersection QoL research may fill a gap in our knowledge of how a life can be lived with different diseases and conditions (Wahl &
Hanestad, 2004). The three levels of QoL will be described in relation to what they mean to the target group of this study.

Figure 1. Main areas within the concept quality of life in terms of health (Wahl & Hanestad, 2004)

4.3.1 The global perspective on QoL

The global perspective can be described as contentment linked to an understanding of what QoL represents. It can be described as the total satisfaction or happiness in life in addition to psychological personality features, motivation, values and preferences.

A definition explains QoL as satisfaction or dissatisfaction with aspect of life that are important for each individual (Ferrans, 1990). Factors like economy, occupation, living accommodation and political condition can have a significant importance in this perspective.

Global single-item measure allows the subject to define the concept in a way that is personally meaningful. It provides a measure that can be responsive to individual differences.

There is considerable disagreement as to whether it is meaningful to ask patients questions such as “Overall, what would you say your QoL has been like during the last week? (Excellent, very good, good, fair, poor, very poor). Some authors argue that responses to these global questions are unreliable and difficult to interpret and that it is better to ask multiple questions about the many aspects of QoL. Responses to the individual questions can be aggregated to form a summary global score. In practice many instruments include at least one global question in addition to a number of multi item scales (Fayers & Machin, 2007).
The last item in the SRI questionnaire is also of global character. That is item 49 “generally I am satisfied with my life”.

QoL questionnaires of global character can in similarity with the general QoL questionnaires be used to compare the healthy in a society against those suffering from a disease “The Quality of life scale” is an example of a questionnaire of global character (Wahl & Hanestad, 2004). This perspective is important for the individual persons, but may be less importance to clinical health practice because global QoL is influenced by other conditions than health.

4.3.2 Health-related quality of life perspective

To obtain limitation relevant to clinical use, the term HRQoL was established as a term in the early eighties, as a mean to separate QoL in general from the aspects that are relevant to health and healthcare (Kaplan & Bush, 1982)). HRQoL can be considered as that part of a persons total QoL that is determined by his or her state of health.

In spite of the wide spread use of the term there is no consensus on how the term should be defined. The concept of HRQoL includes the World Health Organization’s broad definition of health. “Health is a state of complete physical, mental, and social wellbeing and not merely the absence of disease and infirmity” (WHO, 1948).

This definition includes health status and well being. It has been argued that health is a status comprising both wellness and illness, which implies that a person can be ill without a disease. On the other hand persons with severe disease and limitation in health function can perceive themselves as well.

There is consensus that HRQoL should be regarded as a multi-dimensional construct that at least includes physical, psychological and social functioning. The definitions usually refer to physical, social and emotional well-being (Fayers & Machin, 2007a; Wahl & Hanestad, 2004).

For the purpose of this study the definition of Leidy is used. It sees HRQoL as the subjective perception of the impact of health status, including, disease and treatment, on physical, psychological and social functioning and well-being (Leidy, Revicki, & Geneste, 1999).

4.3.2.1 Measuring HRQoL

General questionnaires have often been used in previous HRQoL studies in patient receiving HMV. General questionnaires intend to measure HRQoL independent of specific diseases. These types of questionnaires underline the meaning of health and assume implicit that experiencing poor health equals poor HRQoL. They can therefore be used independently of
any condition, diagnosis, population and intervention. As respiratory and NMD diseases may lead to inability to undertake everyday and social activities requiring exertion, generic scales can generate useful data. Several studies using generic scales have been reviewed (Maille, Kaptein, de Haes, & Everaerd, 1996). One of the most frequent used general scales to measure HRQoL in respiratory patients are Sickness Impact Profile (SIP) (Bergner, Bobbitt, Carter, & Gilson, 1981). Other questionnaires are The Quality of Well-Being Scale (QWBS) (Kaplan, Atkins, & Timms, 1984), The Nottingham Health Profile (NHP) (Hunt, McEwen, & McKenna, 1986) and the SF-36 (McHorney, Ware, & Raczek, 1993; J. E. Ware, 1997, 2008).

There is concern that generic measures may not be sufficiently sensitive. This was illustrated by Schrier who found no correlation between lung function tests and SIP scores (Schrier, Dekker, Kaptein, & Dijkman, 1990). Neither global nor general QoL questionnaires are significantly sensitive in finding changes as a result of treatment or disease (Bowling, 2001). To measure changes in a person's HRQoL related to a disease or a condition, specific questionnaires are needed.

4.3.3 Disease or condition specific QoL perspectives

The disease specific perspective of QoL can be described as the experience of special symptoms and difficulties related to the disease. Disease specific questionnaires give a more detailed description and interpretation of how a limited group of patients experience their own QoL. The focus is kept on how the disease or condition in particular affects the life of the one that is affected. These questionnaires are sensitive to change and will be able to capture the difference between different groups of patients (Fayers & Machin, 2007; Wahl & Hanestad, 2004).

4.3.3.1 Measuring HRQoL in relation to specific diseases and conditions

In some of the studies concerning QoL in patient receiving HMV disease specific questionnaire are used. Guatt’s McMaster Chronic respiratory questionnaire (CRQ) was designed as an outcome measure for people with respiratory disease (Wijkstra, Lacasse, Guyatt, & Goldstein, 2002). The St Georges Respiratory Questionnaire (SGRQ) is a self administered questionnaire which measures impaired health in people with diseases that lead to airway obstruction Carone et al., 2001). Chronic disease assessment tool (CDAT) contain two parts, part 1 contain 106 self reported items in six scales. Part 2 is a clinical section for the recording of the results of pulmonary function test and severity of disease (Moody, 1990).
Studies show that these instruments are not sufficiently sensitive in measuring HRQoL in patients receiving HMV (Windisch, Budweiser, Heinemann, Pfeifer, & Rzehak, 2008; Windisch et al., 2003b). Patients with severe respiratory insufficiency failure can have some of the same symptoms as patients with respiratory disease, but their condition is characterized by more severity in symptoms.

Dyspnoea is the clinical term for shortness of breath. It is probably the most disabling and distressing symptom of lung diseases leading to much anxiety (Bowling, 2001). Also in respiratory failure caused by other diseases, dyspnoea is an important symptom. The patient’s experience of dyspnoea is very relevant and often used as a subjective measure of the patient’s condition. There are a lot of instruments for measuring dyspnea, “Feinstein Index of dyspnea,” “Fletcher Scale,” “MRC Scale,” “American Thoracic Society respiratory questionnaire and grade of breathlessness scale,” “American Lung Association severity of disability,” “Horsley respiratory symptoms questionnaire,” “Severity of symptoms visual analogue scale,” Oxygen-cost diagram, the 6- and 12-minute walking test and stair climbing, “The Borg Ratio perceived scale,” “Mahler baseline and transition dyspnoea Index” (Bowling, 2001). The comprehensive number of instruments also indicates that dyspnoea is an important measurement, but it also shows how difficult it is to choose one instrument and also that it is difficult to compare the result of assessing dyspnea with different instruments.

About one of five HMV users in Norway have a neuromuscular condition. Many of the specific problems and symptoms related to their neurological diseases will not be assessed when using a respiratory disease-specific questionnaire. Within neurology, there are many different diseases that can lead to CRF and need for HMV. Disease-specific questionnaires are developed for many of these different conditions, for example the Barthel Index (Bowling, 2001) and ALS Functional Rating Scale (ALSFRS) (Clarke, Hickey, O'Boyle, & Hardiman, 2001).

These questionnaires do not imply the aspect caused by breathlessness and CRF that can characterize the HMV users. An appropriate specific questionnaire used to measure HRQoL in these patients has to include all these aspects and also has to be sufficiently sensitive and responsive to changes in the patient’s condition concerning dyspnoea and other symptoms caused by their condition.
4.4. Response shift

After describing the different levels of QoL it is important to be aware of a phenomenon called response shift. People with serious illness can have a change in self-evaluation. This entails a change in measuring standard (who to compare to), values (what is important in the individual's life) and redefinition of their situation in life. Many studies show that getting a disease can lead to changes in how one defines QoL. People with life-threatening diseases can therefore be content with their lives, even though they may be feeling uncomfortable and suffering from pain. Many people in difficult circumstances show a capacity to adjust themselves to their new situation and find new meaning in life. It is a result of changes in internal standards, values and understanding of QoL, also called conceptualization (Fayers & Machin, 2007a; Wahl & Hanestad, 2004). This phenomenon can help explain findings from studies which show that people can experience a QoL which is incongruent with apparent observable facts (Richard & Folkman, 2000).

4.5 The choice of HRQoL questionnaire used in the present study

The need for an appropriate questionnaire to measure HRQoL in patients receiving HMV implies the need for a condition-specific questionnaire. Disease- or condition-specific questionnaires are postulated to be more sensitive to changes than general questionnaires. Therefore they are more appropriate when specific therapeutic interventions are being evaluated because they detect differences and changes that arise as a consequence of treatment (Fayers & Machin, 2007). General questionnaires have often been used in previous HRQoL studies in patients receiving HMV. These generic instruments may not be sufficiently sensitive for use in research in this group.

In some studies questionnaire which is specific for only one of the diseases in the HMV population are used. The Norwegian registry of HMV users shows that there are 24 different diagnoses in the adult HMV population. This heterogeneity makes a special challenges to assess HRQoL in this group. Reviewing previous research in HMV also shows how difficult it is to interpretate and compare results in this field because of the extensive use of different instruments.

Therefore, the choice of instrument should be based on a careful consideration of its psychometric properties. A psychometrically strong instrument has to meet several demands and several properties have to be fulfilled for clinical usefulness.
In the present study taking a clinical perspective to HMV patients and their experiences of HRQoL all levels of understanding QoL are relevant, but based on the definition of Leidly especially HRQoL linked to the consequences of treatment are important. When reviewing previous research, two possible questionnaires especially developed for measuring QoL in HMV patients were found, the SRI and the Maugeri Foundation Respiratory Failure Questionnaire (MRF-28). Both were psychometrically tested but neither of them were translated to Norwegian.

It is preferable to use a questionnaire which has already been psychometrically tested. The MRF-28 is a 28-item disease-specific HRQL questionnaire for patients with CRF, either obstructive or restrictive (Carone et al., 1999). It is self-administered, and easy to complete all items being answered as either 'true' or 'false'. It includes 5 questions related to marital and social status, 28 questions with 3 sub scores ('daily activity' based on 11 items, 'cognitive function' based on 4 items, and 'invalidity' based on 5 items) (Janssens et al., 2004).

SRI is a multidimensional condition-specific questionnaire especially developed to measure HRQL in patients receiving HMV. It contains seven domains that cover the most important issue of HRQoL in these patients. This multi dimensional approach is not present in the MRF-28 which does not include psychological aspects of life. The SRI is therefore preferred in this study in favour of MRF-28.

The approach of the team that developed the SRI seems to be very systematic and well documented. In the process of developing SRI they interviewed patients receiving HMV and asked for their experiences in living with HMV. The description of the developing and production process provided information which is fundamental in deciding whether to adapt an existing instrument and which instrument to adapt. The validation of both the German and the Spanish version of SRI gave very good results. (Lopez-Campos et al., 2008; Windisch et al., 2003a, 2003b).

4.6 Translation and adaptation of QoL questionnaire

Translating a questionnaire is more cost-effective than making a new questionnaire, other advantages of using the same questionnaire in several countries is making it possible to do cross-cultural multi-centre studies and compare results from different countries (Skevington, 2002). However, when a questionnaires is translated for use in Norway, it have to be adapted to Norwegian conditions and also be tested for validity and reliability in Norway (Wahl & Hanestad, 2004).
4.6.1 Translation process
The process of translating and adapting a questionnaire has been described by several (Guillemin, Bombardier, & Beaton, 1993; Herdman, Fox-Rushby, & Badia, 1997, 1998; Kvamme, 1998; Wahl & Hanestad, 2004). The first step of the translation process is always to contact the author of the original questionnaire and ask for his or her permission to do the translation.

The sequential procedure for translating is the most common. It includes translation from the original language to the new language and then translating back to the original language. The procedure can be divided into six steps. One: Translation from original language to the new preferred language. Two: Translation back to the original language. Three: Consensus meeting. Four: Pre-testing in a group of professionals Five: Pre-testing in a group of persons from the target group. Six: Psychometric testing (Wahl & Hanestad, 2004).

Translating from original language to the preferred language
Usually at least two different translations are made, by two different translators. Licensed translators is to prefer. It may be an advantage if their social background is different because it might result in a more varied and nuanced language. They have to be familiar with the intention of the questionnaire. An additional benefit may be some knowledge of the subject, although technical terms must be avoided. The translators must not cooperate at this stage of the process; the translations must be made individually (Wahl & Hanestad, 2004).

Translating back to the original language
This must be done by new translators There are different opinions about whether the translators should have any knowledge about the aim of the translation but they must work independently. The number of translators must be the same as the number of translators who performed the original translations (Wahl & Hanestad, 2004). The retranslation is an evaluation of the quality of the original translation and can be considered the golden standard of detecting any flaws, mistakes or shortcomings (Leplege, 1995).

Consensus group
A group of different experts compares and discusses the different versions of the translated questionnaire. The aim is to find the most appropriate sentences and to secure equivalence in the understanding of expressions between the translated and the original version. Short
sentences with an active language and unambiguous denotations are preferred (Guillemin, Bombardier, & Beaton, 1993). To catch the meaning of the questions it may be helpful to communicate with the author of the questionnaire. The result from the consensus group is a version of the questionnaire intended for further testing (Wahl & Hanestad, 2004).

**Pre-testing in a group of professionals**
At first the questionnaire should be tested in a group of professionals to judge the adequacy/relevance of the content and find the best formulation of the items. The professionals ought to have comprehensive knowledge of the current field and also different kind of professionals should be represented to ensure the breadth of knowledge and experience (Wahl & Hanestad, 2004).

**Pre-testing in the target group**
Pre-testing must be conducted in a group of patients belonging to the target group of the questionnaire as well. The chosen group should be as representative of the target group as possible (Leplege & Rude, 1995). The purpose of this is to detect if some of the questions may be confusing, hard to understand, irritating or even offensive to the subject (Fayers, 2007; Guillemin, 1993; Kvamme, 1998).

4.6.1.1 **Criteria for cross-cultural adapting**

To achieve equivalence between the original and the translated questionnaires the following requirements must be met:

*Semantic equivalence* implies a similar meaning of the entire sentence and not a word by word translation.

*Context equivalence* is present when each question describes a phenomenon which is relevant to both cultures.

*Concept equivalence* signifies that the idea and experience of the question is valid to the target group of both cultures.

*Technical equivalence* refers to how the administration method of the questionnaire may affect the results of the questioning. The translated questionnaire and the original must have the same amount of questions, similar scores, and weighting of each answer.

*Criteria equivalence* encompasses the questionnaire’s relation to external criteria. It should be able to discriminate between different subgroups in the target group. To ensure equivalence between the original questionnaire and the translated, the terms and the contain of the
questionnaires must be identical the both of them (Guillemin, 1993; Herdman, 1997; Herdman, 1998; Kvanme, 1998; Wahl, 2004).

4.6.2 Psychometric testing of a questionnaire

A questionnaire developed and tested in one country cannot merely be translated and used in a new version in a foreign country. QoL questionnaires measure subjective and cultural relations and it is therefore necessary to do a psychometric testing of a new version of the questionnaire. The procedure can be looked upon as an evaluation of the translation. It is essential to be as certain as possible that your questionnaire measures what it is supposed to measure and that it is measured in a secure and reproducible way (Bowling, 2005; Streiner & Norman, 2003).

The selected test group should be as representative as possible in relation to the target population (Leplege & Rude, 1995). The size of the selection should also be sufficient in order to make confident generalisations from the answers and further to the entire target population.

A lack of response to a question may have several reasons. For instance a person may forget or do not wish to answer the question. This may be caused by imprecise or offensive questions or that the structure of the questionnaire is unclear. A lack of coherence between question and alternative answers can also be a cause of failure to respond. In addition the questions put forward might not be appropriate for parts or subgroups of the target group. Too many questions, making the questionnaire too comprehensive and time consuming to fill out, could cause the respondent to drop some of the answers.

Consequently, recording and reporting the number of unanswered questions is necessary (Fayers & Machin, 2007). When a person refrains from answering questions in a questionnaire the normal approach is to substitute it with the average value for these people’s answers on a scale. This is applicable if a minimum of 50% of the questions have been answered. The dispersion of answers should also be evaluated in relation to stocking of maximum and minimum scores also called the “floor” and “ceiling” effect. This can lead to a decreased sensitivity and responsively of the questionnaire (Streiner & Norman, 2003).

4.6.2.1 Reliability

The reliability of an instrument indicates how free it is from random error. Reliability refers to the accuracy and consistency of information obtained in a study. The term is often associated
with the methods used to measure research variables. The concept is also important in the analysis of statistical results. It refers to the probability that the results accurately reflect the outcome of a broader group of participations than the actual study has investigated.

Assessment of the stability of an instrument for research involves procedures that evaluate test-retest reliability. It can be found by repeating the measuring within the same group of respondents twice. The comparison is performed statistically by computing a reliability coefficient. A high reliability coefficient is an expression of acceptable variation and repeatability and signifies that the result is not random or elusive (Polit & Beck, 2004).

Internal reliability uses item correlation to assess the homogeneity of multi-item scales. The internal consistence of a questionnaire is a statistical expression for the degree to which the different questions measure parts of the same ability. The most common statistical method for measuring internal consistence is Cronbachs alpha. The range of values is from 0 to +1. Coefficients above 0.70 are generally regarded as acceptable (Fayers & Machin, 2007; Polit & Beck, 2004).

### 4.6.2.2 Validity

Validation of an instrument is a process of determining whether it measures what is intended, and that it is useful for its intended purpose. For example, to what extent is it reasonable to claim that a QoL questionnaire really is assessing QoL? The validation process consists of a number of stages, in which one tries to collect convincing evidence that the instrument produces useful measurements reflecting the respondent’s QoL. The validity can be affected by random and systematic measuring mistakes. Reliability is therefore also an indicator for validity (Brink, 1998). Validity can be subdivided into three main aspects: Content validity, Criterion validity and Construct (Fayers & Machin, 2007).
Content validity
Content validity refers to the process of developing a questionnaire. Are the questions sensible and do they adequately and sufficiently cover the research question? Content validity includes a critical examination of the basic structure of the questionnaire, the reasons why the questionnaire was developed and how it fits the specific research situation.

Face validity or immediate validity is the immediate evaluation of how the questionnaire seems to cover the intended topics clearly and unequivocally. This normally includes a subjective evaluation of experts after the questionnaire is developed. Content validity is taken care of by including a wide range of experts in the developing process (Fayers & Machin, 2007). One can roughly say that QoL embraces physical, social and mental relations. A questionnaire which does not include these aspects cannot be characterised as a QoL questionnaire (Wahl & Hanestad, 2004). There will always be room for discussion whether some items should be included or other removed (Streiner & Norman, 2003).

Criterion validity
Criterion validity involves assessing an instrument against the true value (Fayers & Machin, 2007; Streiner & Norman, 2003). The “true” value in quality of life measurements is not easy to define because there are actual no correct truth about what is QoL. In QoL research try to make probable the best way to seek and define the concept.

Criterion validity can be divided into concurrent validity and predictive validity.

Concurrent validity The most common way to evaluate concurrent validity is to compare the new questionnaire with one or several well-established questionnaires. One example of a well-established questionnaire is SF-36. Criteria validity is confirmed if there is a good correlation between the scores of the two questionnaires.

Predictive validity: The ability of an instrument to predict future, health or sickness or other test results. A prospective longitudinal study will be able to show the questionnaires capability to find the expected situations and concurrancies of the future (Fayers & Machin, 2007).

Construct validity
The construct validity is one of the most important characteristics of a measuring instrument. It is an assessment of the degree to which an instrument measures the construct that it was meant to measure. It involves first forming a hypothetical model, describing the constructs
being assessed and postulating their relationship. Data are than collected, and an assessment is made as to the degree to which these relationships are confirmed. If the results confirm prior expectations about the construct, the implication is that the instrument may be valid and that we may therefore use it to make inferences about patients. Construct validity can be divided into several groups.

Known-groups validation is a simpler form of construct validity. This is based on the principle that certain specified groups of patients may be anticipated to score different from others. The instrument should be sensitive to these differences. A questionnaire that is not sensitive for these differences will not be clinical useful.

Convergent validity is an expression for a dimension with QoL correlates with another dimension. Evaluation of convergent validity includes a preaching of the weakest and strongest coherences between the questionnaires different dimensions and the testing if these are empirically true. Convergent validity is often evaluated together with discriminating validity.

Discriminant validity is based on the presumption that some of the dimensions are anticipated to have low degree coherence and the experienced coherence is low. A relation that is to be aware of is that apparently correlation between to variables in reality is linked with a third variable, spurious correlation.

Sensitivity and responsiveness
Sensitivity and responsiveness are closely linked abilities that describe the questionnaire’s capability to find differences. Sensitivity is the ability to detect differences between patients and groups. Responsiveness is closely related to sensitivity but relates to change within patients. If the patient’s health changes over time the instrument have to detect it to be responsive (Fayers & Machin, 2007).

5. Previous research in Health related Quality of life in patient receiving HMV

Search for previous studies in HRQoL in HMV was performed in electronic databases including MEDLINE, EMBASE, CINAHL, British Nursing Index, Cochrane Library and European Respiratory Journal. Search terms employed were, “health related quality of life” and “home mechanical ventilation” and “not children”. Reference lists of all articles identified for inclusion were manually screened to identify any additional studies. Only studies reported in English were included. Titles and abstracts of all published reports were
identified through the electronic search. The search gave 27 hits. Additional searches were done using the words: “non invasive ventilation” or “bilevel positive airway pressure” and the different main diagnosis.

Reviewing previous research in HRQoL in HMV shows that different methods and instrument have been used. as showed in the bx two and three. Quantitative methods are the most common, but qualitative studies have also been published. The randomized controlled trials regarding treatment effects of HMV are showed in box 2. Some results and conclusions have been criticized on account of methodological issues, for not choosing or enrolling patients into the study group and control group by random (Duiverman, Struik, & Wijkstra, 2008; Wijkstra, 2003). HRQoL is one out of several outcomes in the evaluation of HMV and although generally considered the most important today, many of the earlier effect studies did not include QoL questionnaire. Often only physiologic outcomes were measured before and after initiation of HMV (Simonds, 2003).

In patients with CRF, the distribution of scores for the MRF-28 has been described as wider than for the SGRQ, suggesting that it may be able to discriminate better between different levels of impaired health status than the SGRQ in this particular group (Janssens et al., 2004). The heterogeneity in the HMV group makes it an challenge to describe and review previous research. A summary of some main features and findings in previous studies on HRQoL in HMV is shown in box 2 and 3. Studies that compare QoL in NIV with tracheotomy are described separately.

**Boks 2 RCT studies**

<table>
<thead>
<tr>
<th>First author</th>
<th>Questionnaire</th>
<th>Diagnosis</th>
<th>Outcome data</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Clini, 2002</td>
<td>SGRQ at baseline and at 24 months MRF-28</td>
<td>COPD</td>
<td>Significant improvement in NIPPV group only</td>
<td></td>
</tr>
<tr>
<td>Meecham Jones, 1995</td>
<td>SGRQ</td>
<td>COPD</td>
<td>QoL significantly better QoL</td>
<td>With HMV and LTOT</td>
</tr>
<tr>
<td>Garrod, 2007</td>
<td>CRQ</td>
<td>COPD</td>
<td>Improvement in QoL and fatigue in the NIPPV group</td>
<td>Greater improvement compared with the exercise only group</td>
</tr>
<tr>
<td>Wijkstra, 2003</td>
<td>COPD</td>
<td>ALS</td>
<td>Contradictory outcome</td>
<td></td>
</tr>
<tr>
<td>Bourke, 2006</td>
<td>SF-36</td>
<td>ALS</td>
<td>Improvement in QoL</td>
<td></td>
</tr>
</tbody>
</table>

*Abbreviations in questionnaires:* The St Georges Respiratory Questionnaire (SGRQ), Giatt’s McMaster Chronic respiratory questionnaire (CRQ), Maugeri Foundation Respiratory Failure Questionnaire (MRF-28),
## Box 3 Review on previous research into QoL in HMV patients  (NON-RCT studies)

<table>
<thead>
<tr>
<th>First author</th>
<th>Questionnaire</th>
<th>Diagnosis</th>
<th>Outcome data</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Domenech-Clar, (2003)</td>
<td>SF-36</td>
<td>Restrictive thoracic diseases</td>
<td>Significant improvement</td>
<td>Follow up study</td>
</tr>
<tr>
<td>Bach ,1995, 2002</td>
<td>A specific overall questionnaire</td>
<td>Poliomyelits</td>
<td>Significantly improvement</td>
<td></td>
</tr>
<tr>
<td>Markstrom, 2002</td>
<td>SIP, HI, SOC</td>
<td>Restrictive thoracic diseases, NMD, scoliosis</td>
<td>Significant improvement in both NIV and tracheostomy</td>
<td>Tracheotomy pt. perceived better health, than NIV</td>
</tr>
<tr>
<td>Simonds,1995, 2007</td>
<td>SF-36</td>
<td>Scoliosis, polio pt., prev. tuberculosis. chest wall and</td>
<td></td>
<td>Sleep quality affect QoL</td>
</tr>
<tr>
<td>Dellborg,(2002)</td>
<td>SIP, HAD, MACL, MRC QLQ-C30</td>
<td>scoliosis, previous polio, tuberculosis NMD</td>
<td>Poor QoL in patient with CAH without treatment</td>
<td></td>
</tr>
<tr>
<td>Dellborg,(2008)</td>
<td>SIP, HAD, MACL, MRC QLQ-C30</td>
<td>NON-COPD</td>
<td>Significant improvement HRQoL</td>
<td>Improvement partic. in condition-specific areas</td>
</tr>
<tr>
<td>Storre (2006)</td>
<td>SRI</td>
<td>OHS</td>
<td>Improved HRQoL</td>
<td></td>
</tr>
<tr>
<td>Kaub-Wittemer, 2003</td>
<td>POMS, MLDL</td>
<td>ALS</td>
<td>Good overall QoL for both NIV and TV patients</td>
<td>High burden for TV caregivers</td>
</tr>
<tr>
<td>Rabkin, 2007</td>
<td>PHQ, BDI-PC, BHS, SAHD, QoL, ALSFRS-R.</td>
<td>ALS</td>
<td>More positive life satisfaction in the HMV group</td>
<td></td>
</tr>
<tr>
<td>Windisch, 2008</td>
<td>SRI</td>
<td>NMD, COPD, OHS, RTD</td>
<td>Improvement in HRQoL</td>
<td></td>
</tr>
<tr>
<td>Toussaint, 2007</td>
<td>DMD</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tsolaki, 2008</td>
<td>SF-36</td>
<td>COPD</td>
<td>Improvement in HRQoL</td>
<td></td>
</tr>
</tbody>
</table>

**Abbreviations in questionnaires**: Health Index (HI), Sense of Coherence (SOC), Sickness Impact Profile (SIP), Hospital Anxiety and Depression scale (HAD), Medical Research Council Questionnaire (MRC), Global Quality of life scale (QLQ-C30), Profile of Mood States (POMS), Munich QoL Dimensions List (MLDL), Patient Health Questionnaire (PHQ), Beck Depression Inventory (BDI-PC), Beck Hopelessness Scale (BHS), Schedule of Attitudes toward Hastened Death (SAHD), QoL enjoyment and Satisfaction Questionnaire, Manne Perceived Partner Support Scale. ALSFRS-R.
5.1 Briefly description of HRQoL studies in different diseases

HRQoL in patients with restrictive disorders and stable NMD

The outcome of long term domiciliary nasal intermittent positive pressure ventilation has been analysed in 180 patients with hypercapnic respiratory failure predominantly due to chest wall restriction, neuromuscular disorders, or chronic obstructive lung disease. Outcome measures were survival, pulmonary function, and health status. QoL was measured with SF-36. The long-term outcome of this group with CRF due to scoliosis, previous poliomyelitis, and chest wall and pulmonary disease secondary to tuberculosis is encouraging. The results of NIPPV in patients with COPD and progressive neuromuscular disorders show benefit in some subgroups (Simonds & Elliott, 1995).

A follow up study including forty-five patients with restrictive respiratory diseases, including thoracic wall diseases (n = 27) and NMD (n = 18), underwent 18 months of HMV treatment. SF-36 were used in before treatment and at 3, 6, 9, 12 and 18 months of follow-up. The study conclude that improved QoL and decreased hospitalizations make home non-invasive mechanical ventilation a useful treatment for patients with restrictive respiratory disorders (Domenech-Clar, Nauffal-Manzur, Perpina-Tordera, Compte-Torrero, & Macian-Gisbert, 2003)

A survey of 395 poliomyelitis HMV patients showed on average good satisfaction of life with a score above 4 (7 was highest degree of satisfaction and 1 equalled poorly satisfied). This included all domains in life except health. The same survey reported a tendency of health caregivers to underestimate the patients QoL and to overestimate the difficulties of HMV (Bach, 2002, 2004).

A study that measured HRQL in patients with chronic alveolar hypoventilation (CAH) before starting home mechanical ventilation. Forty-four patients with CAH due to previous polio, scoliosis, healed pulmonary tuberculosis or neuromuscular disease answered a battery of condition specific and generic (Sickness Impact Profile, Hospital Anxiety and Depression scale, Mood Adjective Check List) self-report questionnaires. Patients with CAH due to NMD or restrictive chest wall disorders had severely impaired HRQoL compared to historical data from a healthy reference population (Dellborg et al., 2002).
A prospective study on the impact of HMV on HRQoL in patient with CAH caused by non-COPD conditions were examined at baseline, after 9 months (n=35) and 8 years (n=11) on NPPV. Instrument used were Sickness Impact Profile (SIP), Hospital Anxiety and Depression scale (HADS), and Global Quality of life scale (QLQ-C30 , Mood Adjective Check List (MACL). The instrument used was a condition- specific questionnaire with 18 items was made of items from three different questionnaires. The conclusion was that NPPV improves HRQoL, particularly in condition-specific areas. Improvements were related to effectiveness in ventilation (Dellborg et al., 2008).

**Duchenne muscular dystrophy (DMD)**

DMD patients consider HMV beneficial for independent living and enhancing their overall HRQoL(Kohler et al., 2005)while HRQoL further decreases without MV (Dellborg et al., 2002). Nocturnal NIPPY reverses symptoms associated with poor sleep quality (Ward, Chatwin, Heather, & Simonds, 2005) and full time HMV reverses symptoms associated with daytime hypoventilation such as dyspnoea (Toussaint, Steens, Wasteels, & Soudon, 2006). Full time NIV is possible with a combination of a nasal mask during the night and a mouthpiece during the day, however tracheotomy may be provided when mechanical techniques of cough-assistance are useless to treat chronic cough insufficiency. The ultimate timing to offer full time ventilation with the most advantageous interface is also lacking in evidence. Future investigations are required to enable clinicians to refine the introduction and long-term follow-up of both nocturnal and full time-assisted ventilation in DMD patients (Toussaint, Chatwin, & Soudon, 2007).

**ALS**

The key aims of HMV are relief of symptom and an improvement in HRQoL, while maintaining patient autonomy. It is established that HMV may extend the life of the patients, however the effect on HRQoL have not been systematically studied and data are mainly derived from uncontrolled series (Simonds, 2007). There are few controlled data in this area and further work on HRQoL in both patient and caregiver in addition to long term outcome is required (Simonds, 2007).

So far there has only been one randomized controlled trial of NIV in ALS. Bourke at al followed 92 patients and randomly allocated 19 to standard care and 22 to NIV when they developed entry criteria of either oropnoe or symptomatic daytime hyperkapni. Patients were reviewed at three month intervals and were assessed for symptoms, lung function and HRQoL.
using the SF-36. The subgroup with better bulbar function showed a survival advantage and improvement in HRQoL score. In the group with poor bulbar function there was no survival advantage, but some HRQoL improvement (Bourke et al., 2006).

A study measured QoL and psychosocial issues in ventilated patients with ALS and their caregivers. Two separate disease-specific questionnaires were developed and used, one for the patient and one for the primary caregiver, the Profile of Mood States (POMS) and the Munich QoL Dimensions List (MLDL). Thirty-two patients were ventilated via NIV, 21 via TV. Following conclusions were reached from this study. The data showed a good overall QoL for both NIV and TV patients, but a very high burden of care for TV caregivers, 30% of who rated their own QoL lower than their patient's QoL. Thus, any assessment of QoL in a home palliative care situation should include the primary caregivers (Kaub-Wittemer, Steinbuchel, Wasner, Laier-Groeneveld, & Borasio, 2003).

A study sought to characterize ALS patients who opted for tracheotomy and HMV and compare them with respect to medical, psychiatric, and psychosocial measures to patients who declined tracheotomy and died. 72 ALS patients were included. The questionnaire used was Patient Health Questionnaire (PHQ), Beck Depression Inventory (BDI-PC), Beck Hopelessness Scale (BHS), Holland Systems of Beliefs Inventory, Schedule of Attitudes toward Hastened Death, (SAHD), QoL enjoyment and Satisfaction Questionnaire. Manne Perceived Partner Support Scale. ALSFRS-R.

Fourteen patients chose HMV; 58 died without HMV At study entry, those who later chose HMV were younger, more had young children, higher education, and higher household incomes on average. Although their physical conditions were similar, they reported higher levels of optimism including belief in imminent cure, and more positive appraisals of their ability to function in daily life, their physical health and overall life satisfaction. At study entry, none who later chose HMV were clinically depressed, compared to 26% of those who later refused HMV. The findings suggest that HMV choice was consistent with a sustained sense that life was worth living in any way possible (Rabkin et al., 2006).

**COPD**

A randomized crossover study was performed to measure the effect of the combination of nasal pressure support ventilation (NIPP) and domiciliary LTOT as compared with LTOT...
alone in stable hypercapnic COPD. Fourteen patients were studied, with values of PaO₂, PaCO₂ and FEV₁ recorded. A 4 wk run-in period was followed by consecutive 3-more periods of: (1) oxygen therapy alone, and (2) oxygen plus NIPP in randomized order. The St. George's Respiratory Questionnaire (SGRQ) was used. The result was that QoL with oxygen plus NIPPY was significantly better than with oxygen alone. NIPP may be a useful addition to LTOT in stable hypercapnic COPD (Meecham Jones, Paul, Jones, & Wedzicha, 1995).

A Cochrain review on the outcome of NIPP ventilation in stable patients with COPD is available. In summary this paper concludes that randomised controlled trials (RCT) with a maximum duration of 3 months have shown contradictory effects on blood gasses, dyspnoea, sleep efficiency and HRQoL (Wijkstra, 2003).

Another review, including randomised controlled trials (RCTs) and non-RCTs involving COPD patients who received NIPPV via nasal, oronasal and/or total face mask interfaces. Questionnaires used were MRF-28, SGRQ: St. George's Respiratory Questionnaire; CRDQ: Chronic Respiratory Disease Questionnaire; Only one non-RCT reported HRQoL as an outcome measure and showed improvement in SGRQ scores. Using the MRF-28, Clini demonstrated improvement from baseline in the bi-level NIPPV with LTOT group at 24 months. The study by Garrod showed a significantly greater improvement in QoL and fatigue component in the bi-level NIPPV with exercise group compared with the exercise only group (Kolodziej, Jensen, Rowe, & Sin, 2007).

A recent study measured the effect of the addition of NIV to optimal treatment for 1 year on the HRQoL of stable hypercapnic COPD patients. NIV was offered to 49 out of 58 initially enrolled consecutive patients, of whom 22 refused NIV and comprised the standard treatment group whereas 27 received NIV. HRQoL was assessed with the SF-36 questionnaire. The NIV group showed a significant improvement in QoL by the third month. Dyspnea and diurnal sleepiness improved significantly. No significant improvements were observed in the control group. Patients on NIV spent less days in the hospital compared to controls (Tsolaki et al., 2008).

Published studies shows that although patient with COPD have difficulties adapting to NIV there is some evidence to suggest that nocturnal hypoventilation may be helpful. Patient motivation is critical and hyperkapni patients are more likely to benefit from NIV. Adequate education and acclimatization to the technique is essential (Simonds, 2007).
A recent study was designed to determine whether the MRF-28 and SRI are reliable and valid HRQoL questionnaires in COPD patients with CRF. The SRI proved greater range of measurements and was recommended by the authors. The emphasis in the MRF-28 is mostly on restrictions on activities of daily living, but it underscores the importance of psychological aspects in these patients. However, the MRF-28 adds the cognition domain which may address prevalent and relevant problems in these patients (Duiverman, Wempe, Bladder, Kerstjens, & Wijkstra, 2008).

Another review aimed to integrate current clinical and path physiological concepts, indications for and the utility of non-invasive HMV and its impact on long-term outcome. It seems important to note that improvements in HRQL occurred in measures specifically designed for patients with CRF such as the MRF-28 and the SRI questionnaire. The reviewers concluded that studies looking at patients with COPD and CRF, show beneficial effects on physiological measures and health-related outcomes such as dyspnea and QoL when high treatment pressures and adequate compliance is ensured (Budweiser, Jorres, & Pfeifer, 2008).

**Obesity hypoventilation syndrome (OHS)**

QoL has also been shown to be significantly impaired in patients with OHS without any treatment compared to normal controls (Hida et al., 2003). The aim in one study was to define the most efficient ventilatory treatment modality for patients with OHS. Patients who did not respond to therapy with continuous positive airway pressure were recruited. The effects of bi-level pressure ventilation (BiPAP) with the spontaneous/timed (S/T) ventilation mode were studied in two groups with or without average volume-assured pressure support (AVAPS). Substantially improved oxygenation, sleep quality, and HRQL were found in both patient groups. AVAPS provided additional benefits in terms of ventilation quality, resulting in a more efficient decrease of \( P_{a}CO_{2} \). However, this did not provide further clinical benefits regarding sleep quality and HRQL (Storre et al., 2006).

**Comparison between invasive and non-invasive HMV**

QoL may be perceived differently in consequence of the interface being used to ventilate the patient. Tracheotomised patients express fewer positive statements than those ventilated non-invasively (Brooks et al., 2004; Goldstein, Psek, & Gort, 1995).
There have been no randomized trial specifically addressing this question however and neither is it likely to be conducted. Patients treated by invasive ventilation are likely to have more advanced disease, greater ventilatory dependency and/or bulbar disease (Simonds, 2007). Although experience and experience indicate that NIV have advantages over non-invasive ventilation (Bach, 1995; Bach, 2002).

In a study intermittent positive pressure ventilation (IPPV) by nasal interface was compared to tracheotomy to ALS patients. A starting point for the researchers was the fact that many ALS patients die of respiratory failure without being adequately informed about the available treatment options, such as HMV which can provide symptomatic relief and prolong survival. In conclusion, HMV with tracheotomy -IPPV is an option for selected ALS patients. Nasal-IPPV provides many advantages and should be offered to all patients. However bulbar impairment may limit the usefulness of this option (Cazzolli & Oppenheimer, 1996).

Evaluation QoL of patients with NMD and skeletal diseases treated with non-invasive and invasive HMV was the specific aim of another study. Three questionnaires were used: the Sickness Impact Profile (SIP), the Health Index (HI), and the Sense of Coherence (SOC) scale. Thirty-three patients had post polio dysfunction, 16 patients a neuromuscular disease, 13 patients with scoliosis, and 29 patients (28%) had various other diagnoses. SIP and HI for the Two Treatment Groups (Tracheotomy vs NIV) were collected. The patients with post polio dysfunction and patients with scoliosis treated with tracheotomy perceived the best health index than those treated with NIV for the same diagnoses (Markstrom et al., 2002). In study by Kaub-Wittermann referred to earlier, the scales used to assess QoL did not show any differences between the NIV and the TV groups. Both the POMS and the MLDL instruments measured very similar values in patient and caregiver groups. In particular, the low levels of self-reported depression and anger for both patients and caregivers is notable. When asked directly, 94% of NIV patients and 81% of TV patients would choose ventilation again and more than 80% would advise other patients to do the same. On the other hand, 97% of the NIV caregivers would advise their patient to choose ventilation again, whereas only 75% of the TV caregivers would do likewise (Kaub-Wittemer, Steinbuchel, Wasner, Laier-Groeneveld, & Borasio, 2003).
Quality of life measured with SRI

A multi-centre study tested the hypothesis that both general and condition-specific aspects of HRQL improve following the institution of HMV. In addition, it was hypothesised that improvements would differ according to the underlying disorder. Both general (SF-36) and disease-specific (SRI) aspects of HRQL improved in patients with chronic alveolar ventilation following the institution of HMV. Changes in HRQL were disease-dependent with regard to the single scales of the SRI and SF-36. In particular, significant improvements in Physical Functioning were evident only in COPD and RTD patients. Importantly, improvements in Psychological Well-Being and Social Functioning after one year of HMV were highly significant in COPD, RTD and OHS patients, but were non significant in NMD patients.. Summary Score of the SRI was significantly more sensitive when compared to the PCS of the SF-36 although both summary measures of the SF-36 improved. In conclusion, this study demonstrated that HMV aimed at maximally improving blood gases - seems capable of substantially improving general and condition-specific aspects of HRQL in patients suffering from CRF. Overall improvements in HRQL are comparable in patients with COPD, restrictive thoracic and neuromuscular disorders. The SRI, which specifically targets the sphere of patients who are dependent on HMV, was superior in detecting changes in HRQL when compared to the general instrument SF-36. This highlights the inalienability of disease- or condition-specific instruments for reliably assessing changes in HRQL in patients receiving HMV (Windisch, 2008).

6. Problem formulation

The aims of the study are as previous introduced:

- To translate and trans-culturally adapt the Severe Respiratory Insufficiency Questionnaire, SRI from German into Norwegian, for both patients receiving ventilation via a mask and for patients receiving ventilation via a tracheotomy

and

- To test the psychometrics properties of the Norwegian version of The Severe Respiratory Insufficiency Questionnaire SRI regarding reliability and validity.

for both patients receiving ventilation via a mask and for patients receiving ventilation via a tracheotomy
The problem formulation implies following research questions for this study:

6.1 Translation and trans-cultural adaptation

6.1.1 Is there equivalence between the Norwegian SRI version and the original German version of SRI, considered by the translators and the professionals in the HMV-team?
6.1.2 Do the professionals in the Home mechanical Ventilation team and the members of the patient organization for HMV users, Respira experience the items in the Norwegian version of SRI as clear, meaningful and understandable and not offensive?

6.2 Psychometrics properties:

6.2.1 Is the reliability in the SRI-N satisfactory in the meaning of:
   - Homogeneity with internal consistency revealed as Cronbach alpha coefficient for non-invasive ventilated patients?
   - Homogeneity with internal consistency revealed as Cronbach alpha coefficient for tracheotomised patients?

6.2.2 Is the criterion validity satisfied by the meaning of correlations between the domains in SRI and the domains in SF-36v2 questionnaire?

6.2.3 Is construct validity satisfied by fulfilling the following formulated and expected hypotheses?

1. Different diagnostic groups will be discriminated by the SRI
2. COPD patients have more respiratory complaints
3. COPD patients are more frequently affected by anxiety and depression than patients without COPD
4. Neuromuscular patients have poorer physical function than other patients
5. Patients receiving HMV most hours of the day have a poorer HRQL
6. Impairment of lung function has an influence on quality of life.
7. Methods

In this chapter methods for translation, adaptation and psychometric testing of the translated questionnaire, SRI-N will be described.

7.1 Design

The study was accomplished in two stages. Firstly the questionnaire Severe Respiratory Insufficiency (SRI) was translated and adapted into Norwegian. Secondly psychometric properties of the SRI were tested in a QoL survey including patients from three counties in Norway.

A survey of this kind does not usually have any means of controlling independent variables and is characterised by non-experimental quantitative type of design. It is designed to obtain information by means of self-report about the prevalence, distribution and interrelation in a population by means of self-reporting (Polit & Beck, 2004).

7.2 The Severe Respiratory Insufficiency Questionnaire, SRI

The Severe Respiratory Insufficiency (SRI) questionnaire was presented in Journal of Clinical Epidemiology in August 2003. It’s aiming is to measure HRQoL in patient with CRF receiving HMV. The author is Wolfram Windisch, a physician specialist in pneumonologist working at the University Hospital of Freiburg, Germany. The SRI questionnaire was originally developed in German language following a comprehensive methodology, and it was validated in a multi-centre study including 226 patients and four different hospitals. SRI is a self-administered questionnaire with 49 items. The questionnaire contains seven HRQoL domains, or subscales: respiratory complaints (RC: eight items), physical functioning (PF: six items), attendant symptoms and sleep (AS: seven items), social relationships (SR: six items), anxiety (AX: five items), psychosocial well-being (WB: nine items), and social functioning (SF: eight items). Each item belongs to only one subscale. The items that belongs to the each scale are shown in box 4.
Box 4, SRI scales and items

<table>
<thead>
<tr>
<th>Scale</th>
<th>Item number</th>
<th>Numbers of item</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respiratory complaints (RC)</td>
<td>2, 5, 12, 19, 22, 24, 25 and 29.</td>
<td>8</td>
</tr>
<tr>
<td>Physical functioning (PF)</td>
<td>1, 16, 32, 33, 41 and 45.</td>
<td>6</td>
</tr>
<tr>
<td>Attendant symptoms and sleep (AS)</td>
<td>6, 9, 11, 14, 17, 18 and 42.</td>
<td>7</td>
</tr>
<tr>
<td>Social relationships (SR)</td>
<td>7, 10, 21, 27, 43 and 46.</td>
<td>6</td>
</tr>
<tr>
<td>Anxiety (AX)</td>
<td>8, 13, 26, 28 and 39.</td>
<td>5</td>
</tr>
<tr>
<td>Psychosocial well-being (WB)</td>
<td>4, 20, 30, 34, 36, 38, 40, 44 and 49</td>
<td>9</td>
</tr>
<tr>
<td>Social functioning (SF)</td>
<td>3, 15, 23, 31, 35, 37, 47 and 48.</td>
<td>8</td>
</tr>
<tr>
<td>Summary scale (SS)</td>
<td>All items</td>
<td>49</td>
</tr>
</tbody>
</table>

The choice of items were based on social, psychological, and physical health domains and open interviews in which patients receiving HMV had given important subjective impressions of their actual QoL.

Physical health was covered by items specifically addressing respiratory complaints, such as dyspnea, but also by items related to impairments in physical functioning, and by items covering conditions that are closely related to severe respiratory problems such as sleep disturbances. The psychological domain of HRQL was covered by items asking for well-being and anxieties. In this context, items asking for anxieties particularly related to respiratory disturbances such as “fear of dyspnea” were included. Social aspects of HRQL were reflected by items referring to limitations in social relationships due to the respiratory disease and by items referred to limitations in social activities. The items were developed by an expert panel (physicians specialized in pulmonology, psychologists specialized in HRQL) to assure face validity. All items were rated in a five-point Likert-scale from “strongly agree” to “strongly disagree” in relation to the statements of the items and according to his/her degree of agreement on an ordinal scale with five degrees. Questions refer to his or her health status during the previous week. The items order was randomized. Higher scores were attributed to a better HRQL. Accordingly, items with low scores indicating a better HRQL were recoded to comply with a homogenous data presentation. The total score for each subscale was calculated after recoding certain items and calculating a percentage. After the item were recoded, punctuation of each scale was obtained by a simple mathematical formula. The final punctuation or summary scale (SS) was obtained by the arithmetic mean of the values of each scale. High SS values (range 0–100) indicate a better HRQL.
In the first validation study of SRI construct validity was confirmed by factor analysis, indicating one summary scale that accounts for 59.8% of the variance. Concurrent validity was confirmed by correlating subscales of the SRI and the SF-36 (0.21<r<0.79). Item-scale correlations revealed a high item discriminant validity. The highest correlation was found between SRI-Psychological Well-Being and Mental Health of the SF-36 (r = 0.79). The lowest correlation was found between SRI-Attendant Symptoms and Sleep and Role-Emotional of the SF-36 (r = 0.21). In addition, different diagnostic groups could be discriminated by the SRI. Here, the overall best HRQL was measured in the following order: patients with kyphoscoliosis, miscellaneous disorders, neuromuscular diseases, post-tuberculosis, and chronic obstructive pulmonary disease (P<.05) (Windisch et al., 2003b).

7.3 Procedure for translation and cultural adoption of SRI into Norwegian

The author of the SRI Wolfram Windisch gave his consent to the Norwegian translation.

Translating: German to Norwegian

Accepted procedures for translation and adaptations of QoL instruments were followed (Guillemin, Bombardier, & Beaton, 1993; Wahl & Hanestad, 2004). At first both a professional translator and a physician specialist in pulmonologist with German as first language translated the SRI into Norwegian. They were informed about the aim of the SRI questionnaire. The translators did not cooperate in this phase and worked separately.

Back –Translation: Norwegian to German

Back translation to German was done by two new different professional translators without any knowledge of QoL and HMV. These also worked independently. The researcher and the translators were in contact by mail and phone.

Consensus groups

A group of health care workers compared the translated version and the original for equivalence. This committee consisted of physicians and nurses experienced in HMV. There was a meeting were the different versions of the SRI were examined concerning equivalence. The back translated version of the questionnaire was also sent to the author of SRI. He commented on each item concerning the equivalence between the original version and the
back translated version. The result was a new Norwegian version of the questionnaire ready for further testing, later called SRI-N.

**Pre-testing**

The border of the Norwegian organization for HMV users, “Respira” was contacted and asked to see if the questionnaire was clear and easy to understand. The HMV users were asked for their overall impression of the questionnaire, whether any items had been difficult to answer, annoying or irritating, and whether they had left items unanswered. Five of the members in “Respira” answered this request.

Respira is an interest organization for respirator users and is affiliated The Norwegian Heart and Lung Patient Organisation (LHL). They work for better the conditions for HMV users.

**7.4 Survey for psychometric testing of the SRI-N questionnaire**

A questionnaire developed and tested in one language cannot merely be translated word for word and be used in another language. QoL questionnaire measures subjective and cultural conditions. Therefore it is necessary to do a psychometric testing of the translated questionnaire. The procedure can be taken as an evaluation of the translation. The aim is to test general user friendliness, reliability, sensitivity, responsiveness and how easily understandable the questionnaire is to the target informants (Fayers & Machin, 2007). One have to be as sure as possible that the questionnaire measures what it is supposed to measure and that the measuring is done in a safe and reproducible way (Bowling, 2001; Streiner & Norman, 2003).

**7.4.1 Data collection**

In the present survey data is collected by using three questionnaires, the translated SRI-N and SF-36v2. A specific questionnaire measuring demographic and specific variables is also used. In addition data is collected from the Norwegian Registry of patients receiving HMV.

- The Norwegian version of SRI–N questionnaire
- SF-36 v2 questionnaire
- Questionnaire with demographic and specific variables
- Norwegian Registry of patients receiving HMV
7.4.1.1. Short Form SF-36v2

For validation purposes, the SF-36v2 was administered together with the SRI. The SF-36 is a 36-item self-report questionnaire measuring physical functioning (10 items), role-physical (4 items), emotional role limitations (4 items), bodily pain (2 items), mental health (5 items), social functioning (2 items), general health (5 items) and vitality (4 items). Which items that belongs to each sub-scale and also which subscale that belongs to each of the two summary scales are shown in box 5.

Box 5. SF36 Health Survey Measurement Model (J. E. Ware et al., 2008)

<table>
<thead>
<tr>
<th>Items</th>
<th>Scale</th>
<th>Summary measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>3a. Vigorous Activities</td>
<td>Physical functioning (PF)</td>
<td>Physical (PCS)</td>
</tr>
<tr>
<td>3b. Moderate Activities</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3c. Lift, Carry Groceries</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3d. Climb Several Flights</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3e. Climb One Flight</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3f. Bend, kneel</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3g. Walk Mile</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3h. Walk Several Hundred Yards</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3i. Walk one Hundred Yards</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3j. Bath, Dress</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4a. Cut Down Time</td>
<td>Role-Physical (RP)</td>
<td></td>
</tr>
<tr>
<td>4b. Accomplished Less</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4c. Limited in Kind</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4d. Had difficulty</td>
<td></td>
<td></td>
</tr>
<tr>
<td>7. Pain-Magnitude</td>
<td>Bodily Pain (BP)</td>
<td></td>
</tr>
<tr>
<td>8. Pain-Interference</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1. EVF Rafting</td>
<td>General Health (GH)</td>
<td></td>
</tr>
<tr>
<td>11a. Sick Easier</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11b. As Healthy</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11c. Health to Get Worse</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11d. Health Excellent</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9a. Full of life</td>
<td>Vitality (VT)</td>
<td>Mental (MCS)</td>
</tr>
<tr>
<td>9b. Nervous</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9c. Down in Dumps</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9d. Peaceful</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9e. Energy</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9f. Downhearted/Depressed</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9g. Worn Out</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9h. Happy</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9i. Tired</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
The SF-36 was developed by Ware et al based on the Medical Outcome Study (MOS), which was based on a multidimensional model of health (J. E. Ware, Jr., 1987). SF-36 is referred to as a generic health measure, and such as is not specific for age-, disease or treatment group. It is translated into many language including to Norwegian, by Loge et al (Loge & Kaasa, 1998). It assesses HRQoL outcomes, namely those outcomes known to be most directly affected by disease and treatment (J. E. Ware, Jr., 2000). The scores in each domain are transformed into a scale from 0 to 100 where higher scores indicate better HRQoL. SF-36 has become the most widely used of all generic health measures. The questionnaires psychometric properties are well documented (J. E. Ware, Jr., 2000).

Although the SF-36 Health Survey proved to be useful for many purposes, 10 year of experience revealed the need and potential for improvements. The result of several revision and evaluation studies led to the development of SF-36 version 2 (SF-36v2). It represent an improved measurement tool that maintains comparability with the original version in terms of purpose, content, scores and the psychometric which it was developed. It can be used across all adult patient and non-patient populations for variety of purposes, such as screening individual patients, monitoring the result of care, comparing the relative burden of disease, and comparing the benefits of different treatments.

Without increasing the number of questions, the SF-36v2 substantially increase the reliability and validity of scores and make a survey easier to understand and complete. Further the norm based scoring (NBS) algorithms make it possible to compare results across both versions of the SF-36. Enhancement of wording and response categories reduced extend of floor and ceiling effecting the role performance health domain. These advances are likely to lead to better precision as well as greater responsiveness in longitudinal studies (J. E. Ware et al., 2008).

The new version is license required and this study has obtained the necessary license from Quality Metric.

7.4.1.2 Sosiodemographic data and specific variables

The participants also received a questionnaire asking for demographic background variables, there were also questions about education in using the ventilator and follow up from the health services. The sosiodemographic data were age and gender and education level (Appendix 8).
7.4.1.3 Norwegian Registry of patients receiving HMV

Clinical data concerning diagnosis and disease severity and performance status were collected from the Norwegian Registry of patients receiving HMV. Inclusion criteria for the registry are all Norwegian patients on long-term ventilation for domiciliary use. Patients on continuous positive airway pressure (CPAP) or ventilators for physiotherapy only were not included. The first patient was registered February 15th 2002.

Clinical data collected to the present study were main diagnosis, the main indication for establishing HMV, duration of HMV (years), type of connection to the ventilator, type of ventilator, additional treatment like connection to oxygen, body mass index, blood gas analysis (PO2, PCO2,) and spirometry values (FVC and FEV1).

7.4.2. Study sample

Study participants consisted of all patients in the Norwegian Medical Centre for Home Mechanical Ventilation registry from three counties in Norway. They were all over 18 years of age. Persons < 18 years were excluded because research in QoL in adolescent and children require particular methods and instruments (Polit & Beck, 2004). Both patients with non-invasive and invasive ventilation were included. HMV ventilation had to be well adapted for at least 3 months. The patients had to be mentally clear and oriented. 214 patients met these criteria.

7.4.3 Procedure

The standardised questionnaires, an information letter and a stamped return envelope were sent by post to the HMV users who met the eligibility criteria. Returning the questionnaire was seen as consent to participate in the study. In the information letter the HMV users were told about the aim of the study and informed that clinical data concerning diagnosis, disease severity and performance status were collected from the Norwegian Register of patients receiving HMV (Appendices 4). After 1 month, a reminder letter together with copies of the questionnaires were sent to the non-responders.

The register was crosschecked with the “Folke registeret “/ “The National Inhabitant Registry” before mailing the questionnaires. One patient had died the same week that the questionnaire were sent to the persons that met the inclusion criteria. Nine patients returned
the questionnaire unanswered because they had stopped using the ventilator. Two patients were not able to answer the questionnaire, as judged by their relatives or carers. Two patients were not possible to locate. One of the patients who had received the questionnaire was under 18 years of age. This brought the number of potential responders down to 199.

It was considered sending the questionnaire by e-mail since some of the HMV users are active on the internet. However, most of the HMV users do not have access to the internet and sending the answers on ordinary e-mail could also have made it difficult to adhere to the rules of confidentiality.

### 7.4.4 Reliability

Assessment of reliability was applied with internal reliability, which is often called internal consistency. It is based on item to item correlations in multi item scales to assess homogeneity (Fayers & Machin, 2007). The internal consistency of the SRI was evaluated for the total sample in each domain separately and in the total of all the domains. Both patients with tracheotomy and non invasive HMV users are included in the study. To ensure the reliability, internal consistency is also evaluated separated for the non-invasive patients and the patients with tracheotomy.

Because of possible of misinterpretation of item 15, chronbach alfa also was measured for SRI-SF domain without item 15. The misinterpretation may be the difference in feeling “bonded to” or “connected to” his / her home.

It would have been desirable to measure the repeatable reliability, by sending out the questionnaire SRI a second time and used a test–retest or inter rater reliability. The reason for not doing this was partly limitation of capacity and resources in a master-study context and because the researcher was afraid to bother the HMV user twice.

### 7.4.5. Validity

In the present study content/ face validity, criterion-related validity and construct validity was assessed (Fayers & Machin, 2007; Streiner & Norman, 2003).

**Content / Face validity:** Members of the Norwegian organization for HMV users, Respira, and professional’s members of the HMV team assessed the SRI-N to evaluate if it was clearly and easy to understand and covered the topic of interest.
**Criterion validity:** The SF-36 questionnaire was used as an “objective gold standard” for criterion validity in the validation study of the original SRI (Windisch et al., 2003b). In the present study SF-36 was administered together with the translated SRI-N. Criterion validity was determined by comparing the Norwegian SRI questionnaire with the SF-36v2 and measure correlation coefficients between the different scales in both questionnaires. In this case, a high correlation between scales studying similar aspects of HRQL in both questionnaires was expected to confirm criterion validity.

**Construct validity:** Known-groups validity, which tests the instrument for discriminatory ability, was chosen for evaluating construct validity. Hypotheses were formulated to see if the questionnaire fulfilled them (Fayers & Machin, 2007; Streiner & Norman, 2003). The formulated hypotheses were based on results from previous studies. Hypothesis 1 is based on the postulate that HRQL is influenced by the underlying disease. (Windisch, 2008; Windisch, Budweiser, Heinemann, Pfeifer, & Rzehak, 2008; Windisch et al., 2003b)

Hypothesis 2 is based on that COPD patients are expected to have more respiratory complains than patients without lung diseases. Hypothesis 3 is formulated because of studies using a questionnaire specifically designed to measure anxiety and depression have shown significantly more frequent affected by anxiety and depression in COPD patients compared to patients with kyphoscoliosis or neuromuscular diseases when being on HMV (Windisch, Petermann, Laier-Groeneveld, Fischer, & Crie, 1997) Accordingly, it is expected that anxiety and impairments in well-being are more frequent reported in patients with COPD when measuring HRQoL with the SRI.

Although the physical functioning of patients with neuromuscular diseases is impaired by respiratory dysfunction, it is mainly impaired by weakness of the limbs. Therefore, limitations in physical functioning were expected to be more severe in this group of patients compared to those patients who do not additionally suffer from weakness of the limbs (Windisch, 2008; Windisch, Budweiser, Heinemann, Pfeifer, & Rzehak, 2008; Windisch, Freidel, Matthys, & Petermann, 2002; Windisch et al., 2003a). These assumptions give hypothesis 4.

Hypothesis 5 is based on the assumption that patients receiving HMV most hours of the day have a poorer HRQL. Earlier studies have shown inconsistent degrees of correlation between physiologic parameters of lung function (FVC and FEV1) and HRQoL (Yessen, 2000). Nevertheless it may seem probably that there is a correlation between lung function and HRQoL in these severe ill patients, and hypothesis 6 is based on these assumptions.
7.5 Statistical analyses

Pre-analysis

In the pre-analysis phase the filled out questionnaires was transferred to computer files in the statistic program SPSS (Statistical, Package of the Social Science) (SPSS, Chicago, IL, USA) version 12.0. The analysis was carried out with the assistance of a statistician. To ensure the process of punching of the information, the files were double-checked in addition to a check-up of every third schema. Missing items in the SF-35 and SRI were treated according to accepted guidelines for SF-36 and SRI. A substitution in terms of an average value of the remaining score values within the domain were done if at least 50 present of the items in the domain were completed (Fayers & Machin, 2007; J. E. Ware et al., 2008; Windisch et al., 2003b). According to guidelines for the original SRI questionnaire these items were recoded: 1, 2, 4, 5, 6, 8, 11, 12, 13, 14, 15, 16, 17, 19, 21, 22, 23, 24, 25, 26, 28, 29, 30, 31, 34, 35, 38, 39, 40, 42, 43, 44, 45, 46, 47, and 48.

Box 6. These items were recoded as followed:

<table>
<thead>
<tr>
<th>Original value</th>
<th>Recoded value</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>5</td>
<td>1</td>
</tr>
</tbody>
</table>

Analysis phase

The first statistical analysis in the present study was to find out if there were differences between the participants and the non-participants. The measurement levels of the different variables are the starting point when choosing a statistic analysis. The parametric tests demand data to be at interval or ratio level, and do often include assumptions about the population (e.g. normally distributed). The non-parametric tests do not have such stringent requirements and do not make assumptions. They are ideal to use when data are on nominal (categorical) and ordinal (ranked) scales.
Independent-samples t-test was used to explore the difference between the participants and the non-participants in continuous variables. Because of the assumptions that the population distribution may not be normally distributed, Mann-Whitney U non-parametric test was also used. For descriptive purposes, the mean ± standard deviation was used for quantitative variables, and the absolute and relative frequencies of each category were used for qualitative ones.

A one way between groups analysis of variance (ANOVA) was conducted to explore the difference between the four main diagnosis groups.

The total sum score in SRI is calculated by the average (SRI-RC, SRI-PF, SRI-AS, SRI-SR, SRI-AX, SRI-WB, SRI-SF). If the results are missing in one of the scales the calculation will not be done.

**Reliability test:** Internal consistency was determined by calculation of Cronbach’s $\alpha$ of each domain, sub-domain and in the total scale score.

**Factor analysis:** In the Spanish translation of SRI a factor analysis was performed using the principal component method. In the present study the principal axis factoring method was used. It takes a large set of variables and looks for a way the data may be “reduced” or summarised using a smaller set of factors or component.

**Validity tests:** In the analysis all the scores in SF-36 and in SRI are recalculated to a scale from 1 to 100. The data were partly normal distributed but there were some ceiling effect, non-parametric tests are therefore used. Criterion validity was determined by Spearman rank correlation for association between health domains in the SRI-N and the SF-36. A $P<0.05$ was considered statistically significant (Pallant, 2007) SPSS.

Construct validity was determined by the non-parametric tests, Mann–Whitney test and the A Krustal- Wallis Test when the variables were on ordinal (ranked) scale.

**Hypothesis testing:** A hypothesis is a statement of the researcher’s expectations of relations between the variables under investigation (Polit & Beck, 2004)

Construct validity was tested by formulating the anticipated and accepted hypotheses. The procedure used in testing hypotheses is based on rules of negative interference. It states that there is no relationship between variables. It is basically a process of disproof or rejection. It cannot be demonstrated directly that the research hypothesis is correct, but it is possible to
show, using theoretical sampling distributions, that the null hypothesis has a high probability of being incorrect (Polit & Beck, 2004).

The null hypothesis for testing the construct validity is that there is no difference in HRQoL between different diagnosis groups or other characteristics in coherence with HMV. In hypothesis testing the researcher can make two types of error. The error variation might be random or/and systematic. The random variation is a threat of the reliability and the systematic variation affects the validity. Type 1 error is to deny null hypotheses when it is correct. Type 2 error is to accept a false null hypothesis. The risk of doing these mistakes is affected by the significance level (Polit & Beck, 2004).

In the hypothesis testing the independent-samples t-test is used to compare the mean scores of different groups. Construct validity was also determined by the Mann–Whitney test. The Krustal- Wallis test is used to compare the score on continuous variable for three or more groups. Qualitative variables were studied using the exact chi-square test and was used to determine whether the categorical variables were related to different diagnosis groups.

7.6 Ethics

The present study has followed the ethical guidelines of the Declaration of Helsinki from 1964, amended in October 2008 (World Medical Association, 2008). The study was approved by the Norwegian Committee of Ethics in Medicine, Region III (Appendix 2) by the Norwegian registry of Data-security (NSD)(Appendix 1).

In the information letter which was sent to the eligible and included HMV users it was informed that they were contacted because they were in the Norwegian Registry of Patients receiving HMV and that data from the registry would be used to get information about treatment, type of ventilator and other equipment used. Further they were informed about the aim of the study and that participation was voluntary and that it would not affect their treatment or follow up from the Hospitals if they did not return the questionnaire. It was informed that this project would be completed in 2008 but the data were preserved for 10 years for a possible follow-up study. The participants were guaranteed confidentiality. They were also informed that if they wanted to ask questions about the study or needed help answering the questions they could contact the project leader by telephone or e-mail.
In studies with humans, it is of crucial importance to avoid, prevent or minimize harm (Polit & Beck, 2004). The questionnaires used in the present study were considered not to cause any harm or mental distress.
8. Results

8.1 Trans-cultural adaptation of the SRI questionnaire

8.1.1 Translation, back translation and the consideration from the consensus group

The translation process revealed four versions of the SRI-N, before agreement upon a common version. There was divergence in translation in item 1, 2, 4, 5, 9, 15, 19, 23, 26, 28, 29, 30, 34, 35, 36, 37, 38, 39, 42, 44, 45, 46, 47, 49 and modifications of wording were made in these item numbers.

Concerning equivalence one word was especially challenging. “Luftnot” (English: Breathlessness) was used in many of the items. Both patients and professionals had the same perception of the meaning, but used different Norwegian word, like “tungpust”, “åndenød”, “dyspnoe”, “kortpusten”, “pustebesvær”, and “pusteplager”. The semantic equivalence is concerned with the meaning across language, and with achieving a similar effect on respondents using different languages. The original author also approved the final version.

8.1.2 Pre-testing

In the pre-testing five of the members of Respira responded that the questionnaire was clear and understandable. They did not think it was too long and the items made sense concerning their lives as HMV users. One of them complained about item 1. He was a wheelchair user and found the item “is it difficult for you to use the stairs “difficult to answer. He also meant that the questionnaire did not approach in a sufficient way the fact that a large number of patient receiving HMV are very disabled. His experience was that the SRI questionnaire was too focused on and addressed to HMV users who mainly have breathing problems and not all the other factors that impact their daily living.

One of the responders from the Respira organization reported that in addition to the SRI questionnaire he would like to have questions which included direct evaluation of the Public Health service, e.g. “Do you get enough help from the Public Health Service?” Do you get the necessary equipment concerning the ventilator and do you get it on time? “

However, finally patients, translators and health care workers reached consensus, and the SRI-N is shown in Appendix A.
8.2 Psychometric testing of the SRI questionnaire

8.2.1. Patient material

131 patients answered the questionnaire, but four of the responders were excluded after it was discovered that they had obstructive sleep apnoea and therefore did not fulfil the criteria for the Norwegian Registry of patients receiving HMV. The study group therefore consists of 127 participants, giving a response rate of 64 %.

Descriptions of participants and non participants

The demographic and clinical characteristics of the participant and non-participants are summarized in table 1. There were no significant differences between participants and non-participants regarding age, sex, different diagnosis and year of HMV. However, an independent sample t-test revealed a significant difference in FVC and FEV1 between responders and non responders, \( p<0,005 \).

| Table1. Demographic and clinical characteristics participants and non participants |
|------------------------------------------|---------|---------|---------|
|                                | Participant | Non participant | \( p \) value |
|                                | N=127 | Mean SD | % | N=66 | Mean SD | % |          |
| Age(years)                      | 61.5 ±15.6 | 58 ±21.27 | P=0.250 |
| Gender (female/male)            | 47/53 | 44/56 | P=0.739 |
| Duration of HMV(years)          | 4.92 ±4.05 | 4.67 ±3.26 | P=0.674 |
| PO2                             | 8.6 ±2.24 | 8.3 ±2.29 | P=0.52 |
| PCO2                            | 7.16 ±2.4 | 7.16 ±2.0 | P=0.993 |
| FVC                             | 2.4 ±1.0 | 1.8 ±0.9 | P<0.005 |
| Percent predicted FVC           | 64.6 ±23.6 | 53.8 ±23.2 | P<0.052 |
| FEV1                            | 1.62 ±0.96 | 1.32 ±0.7 | P<0.043 |
| NMD                             | 35 % | 33 % |
| COPD                            | 19 % | 15 % |
| OHS                             | 29 % | 33 % |
| Other diagnosis                 | 15 % | 18 % |
Abbreviations: HMV, home mechanical ventilation; FVC, forced vital capacity; FEV$_1$, forced expiratory volume in one second; NMD, neuromuscular disease; OHS, obesity-hypoventilation syndrome; COPD, chronic obstructive pulmonary disease.

Descriptions of non-invasive HMV patients and patients receiving ventilation via a tracheotomy

115 of the patients in the study group were ventilated in a non-invasive mode with a facial or nasal mask, and 11 were ventilated via a tracheotomy. The independent T-test did not show any significant difference in the non-invasive and the tracheotomised participants regarding age, sex, Po2, FEV1 and VK (table 2).

Table 2 Description of non-invasive HMV patients and patients receiving ventilation via a tracheotomy

<table>
<thead>
<tr>
<th></th>
<th>Non Invasive N=115</th>
<th>Patients with tracheotomy N=11</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean   SD   %</td>
<td>Mean   SD   %</td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td>62,4 ±15,1 53,9 ±19,6</td>
<td>P=0,101</td>
<td></td>
</tr>
<tr>
<td>FEV1</td>
<td>1,6 ±0,98 1,5 ±0,80 *</td>
<td>P=0,79</td>
<td></td>
</tr>
<tr>
<td>FVC</td>
<td>2,4 ±1,0 2,0 ±1,1 *</td>
<td>P=0,52</td>
<td></td>
</tr>
<tr>
<td>Po2</td>
<td>8,5 ±2,3 9,6 ±1,9</td>
<td>P=0,28</td>
<td></td>
</tr>
<tr>
<td>HMV hour a day</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5-8 hr/ day</td>
<td>52% 9,1%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>8-12 hr/day</td>
<td>38% 18,2%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>12-18 hr/day</td>
<td>4,3 % 27,3%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>18-24 hr/day</td>
<td>5,2 % 45,5%</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* FVC and FEV1 were only measured in five patients with non invasive ventilation.

Abbreviations: HMV, home mechanical ventilation; FVC, forced vital capacity; FEV$_1$, forced expiratory volume in one second;

The study group was divided into 4 main groups. In the group of NMD patients there were 10 patients receiving ventilation via a tracheotomy. This group also received HMV most hours of a day. In the COPD and OHS groups there were none with tracheotomy. In the group of other diagnosis there was one patient receiving ventilation via a tracheotomy.
### Table 3  Clinical characteristics of the patient sample

<table>
<thead>
<tr>
<th></th>
<th>NMD N=45</th>
<th>COPD N=25</th>
<th>OHS N=38</th>
<th>Other N= 19</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>SD</td>
<td>%</td>
<td>Mean</td>
</tr>
<tr>
<td>HMV hours a day</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5-8 hr/ day</td>
<td>51</td>
<td>28</td>
<td>47</td>
<td>63</td>
</tr>
<tr>
<td>8-12 hr/day</td>
<td>30</td>
<td>52</td>
<td>38</td>
<td>31</td>
</tr>
<tr>
<td>12-18 hr/day</td>
<td>2</td>
<td>8</td>
<td>8</td>
<td>5</td>
</tr>
<tr>
<td>18-24 hr/day</td>
<td>16</td>
<td>12</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td>VC (%)</td>
<td>64,1 ±28,2</td>
<td>58,3 ±19,1</td>
<td>70,1 ±17,3</td>
<td>61,7 ±33,7</td>
</tr>
<tr>
<td>FEV₁</td>
<td>1,4 ±0,8</td>
<td>1,1 ±0,67</td>
<td>2,1 ±1</td>
<td>1,6 ±1,1</td>
</tr>
<tr>
<td>FEV₁ (%)</td>
<td>55,9 ±26,4</td>
<td>36,2 ±18,9</td>
<td>63,0 ±23,9</td>
<td>58,1 ±29,0</td>
</tr>
<tr>
<td>pO₂</td>
<td>9,9 ±1,8</td>
<td>7,0 ±2,1</td>
<td>7,9 ±1,9</td>
<td>9,7 ±2,4</td>
</tr>
<tr>
<td>Tracheotomy</td>
<td>22</td>
<td>0</td>
<td>0</td>
<td>5</td>
</tr>
<tr>
<td>Non invasiv</td>
<td>78</td>
<td>100</td>
<td>100</td>
<td>95</td>
</tr>
</tbody>
</table>

**Abbreviations:** NMD, neuromuscular disease; OHS, obesity-hypoventilation syndrome; COPD, chronic obstructive pulmonary disease; HMV, home mechanical ventilation; FVC, forced vital capacity; FEV₁, forced expiratory volume in one second.

---

**The distribution of the sample in the different SRI domain**

The distribution of the sample in the different SRI domains do not seem to be stringent fully normally distributed. The graph in figure 2 is showing the distribution in SRI respiratory component (RC) reveals that there are some ceiling effects in the result.
8.2.2 Reliability

As shown in table 4 the Cronbach alpha coefficient obtained in each domain varied from 0.76 to 0.88. Because of possible misinterpretation of item 15 chronbach alfa also was applied in SF domain without item 15. The result was then 0.80.

Table 4. Internal consistency for SRI- N for the total sample

<table>
<thead>
<tr>
<th>Scale</th>
<th>Number of item</th>
<th>Cronbach alpha</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respiratory complaints (RC)</td>
<td>8</td>
<td>0.81</td>
</tr>
<tr>
<td>Physical functioning (PF)</td>
<td>6</td>
<td>0.76</td>
</tr>
<tr>
<td>Attendant symptoms and sleep (AS)</td>
<td>7</td>
<td>0.68</td>
</tr>
<tr>
<td>Social relationships (SR)</td>
<td>6</td>
<td>0.82</td>
</tr>
<tr>
<td>Anxiety (AX)</td>
<td>5</td>
<td>0.81</td>
</tr>
<tr>
<td>Psychosocial well-being (WB)</td>
<td>9</td>
<td>0.88</td>
</tr>
<tr>
<td>Social functioning (SF)</td>
<td>8</td>
<td>0.79</td>
</tr>
<tr>
<td>Summary scale (SS)</td>
<td>49</td>
<td>0.94</td>
</tr>
</tbody>
</table>
**Table 5. Cronbach alpha coefficient for the NIV patients, patients with tracheotomy were excluded**

<table>
<thead>
<tr>
<th>Scale</th>
<th>Number of item</th>
<th>Cronbach alpha</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respiratory complaints (RC)</td>
<td>8</td>
<td>0.81</td>
</tr>
<tr>
<td>Physical functioning (PF)</td>
<td>6</td>
<td>0.77</td>
</tr>
<tr>
<td>Attendant symptoms and sleep (AS)</td>
<td>7</td>
<td>0.66</td>
</tr>
<tr>
<td>Social relationships (SR)</td>
<td>6</td>
<td>0.83</td>
</tr>
<tr>
<td>Anxiety (AX)</td>
<td>5</td>
<td>0.83</td>
</tr>
<tr>
<td>Psychosocial well-being (WB)</td>
<td>9</td>
<td>0.89</td>
</tr>
<tr>
<td>Social functioning (SF)</td>
<td>8</td>
<td>0.82</td>
</tr>
<tr>
<td>Summary scale (SS)</td>
<td>49</td>
<td>0.89</td>
</tr>
</tbody>
</table>

**Table 6. Cronbach alpha coefficient for the patients with tracheotomy, patient ventilated with NIV were excluded**

<table>
<thead>
<tr>
<th>Scale</th>
<th>Number of item</th>
<th>Cronbach alpha</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respiratory complaints (RC)</td>
<td>8</td>
<td>0.84</td>
</tr>
<tr>
<td>Physical functioning (PF)</td>
<td>6</td>
<td>0.56</td>
</tr>
<tr>
<td>Attendant symptoms and sleep (AS)</td>
<td>7</td>
<td>0.61</td>
</tr>
<tr>
<td>Social relationships (SR)</td>
<td>6</td>
<td>0.52</td>
</tr>
<tr>
<td>Anxiety (AX)</td>
<td>5</td>
<td>0.69</td>
</tr>
<tr>
<td>Psychosocial well-being (WB)</td>
<td>9</td>
<td>0.82</td>
</tr>
<tr>
<td>Social functioning (SF)</td>
<td>8</td>
<td>0.55</td>
</tr>
<tr>
<td>Summary scale (SS)</td>
<td>49</td>
<td>0.89</td>
</tr>
</tbody>
</table>
8.2.3 Validity

8.2.3.1 Face validity

Translators, HMV users, members of the HMV team and the author of the original questionnaire made a consensus upon a final version of the SRI-N. The pre-testing confirms the SRI-N to be clear and understandable and covering the topic of interest.

8.2.3.2 Structural validity

Explorative factor analysis for SRI, principal axis factoring, varimax-rotasjon: explained 67% of the variation of the questionnaire and revealed 13 factors. When factor analysis were applied with 7 factors the result explained 55% of the variation of the questionnaire.

Figur 3. Plot of the factor analysis.
8.2.3.3 Criterion validity

Criterion validity for the total sample

The correlation matrix for SRI-N and SF-36v2 for the total study group is showed in Table 7. Spearman rank correlation revealed the best correlation between physical functioning (PF) in both SRI-N and SF-36v2 (r=0.729; p<0.001) and between SRI Well-being (WB) and SF-36v2 vitality (VT) scales (r=0.72; p<0.001) Also between SRI-N WB and SF-36v2 MHC (r=0.714; p<0.001).

Table7. The correlation matrix for SRI-N domains and SF-36 v2- domains for the total study group.

<table>
<thead>
<tr>
<th>SRI-N</th>
<th>PF</th>
<th>RP</th>
<th>BP</th>
<th>GH</th>
<th>VT</th>
<th>SF</th>
<th>RE</th>
<th>MH</th>
<th>PHC</th>
<th>MHC</th>
</tr>
</thead>
<tbody>
<tr>
<td>RC</td>
<td>0.378</td>
<td>0.520</td>
<td>0.362</td>
<td>0.633</td>
<td>0.521</td>
<td>0.515</td>
<td>0.400</td>
<td>0.286</td>
<td>0.527</td>
<td>0.396</td>
</tr>
<tr>
<td>PF</td>
<td>0.729</td>
<td>0.661</td>
<td>0.246</td>
<td>0.524</td>
<td>0.345</td>
<td>0.398</td>
<td>0.430</td>
<td>0.270</td>
<td>0.608</td>
<td>0.290</td>
</tr>
<tr>
<td>AS</td>
<td>0.172</td>
<td>0.206</td>
<td>0.494</td>
<td>0.359</td>
<td>0.436</td>
<td>0.330</td>
<td>0.252</td>
<td>0.323</td>
<td>0.339</td>
<td>0.272</td>
</tr>
<tr>
<td>SR</td>
<td>0.272</td>
<td>0.430</td>
<td>0.481</td>
<td>0.475</td>
<td>0.579</td>
<td>0.664</td>
<td>0.418</td>
<td>0.523</td>
<td>0.465</td>
<td>0.582</td>
</tr>
<tr>
<td>AX</td>
<td>0.297</td>
<td>0.408</td>
<td>0.449</td>
<td>0.499</td>
<td>0.436</td>
<td>0.582</td>
<td>0.439</td>
<td>0.494</td>
<td>0.421</td>
<td>0.548</td>
</tr>
<tr>
<td>WB</td>
<td>0.281</td>
<td>0.480</td>
<td>0.543</td>
<td>0.629</td>
<td>0.720</td>
<td>0.695</td>
<td>0.580</td>
<td>0.637</td>
<td>0.430</td>
<td>0.714</td>
</tr>
<tr>
<td>SF</td>
<td>0.417</td>
<td>0.613</td>
<td>0.449</td>
<td>0.589</td>
<td>0.518</td>
<td>0.656</td>
<td>0.446</td>
<td>0.373</td>
<td>0.587</td>
<td>0.489</td>
</tr>
<tr>
<td>SS</td>
<td>0.452</td>
<td>0.617</td>
<td>0.578</td>
<td>0.702</td>
<td>0.645</td>
<td>0.736</td>
<td>0.560</td>
<td>0.537</td>
<td>0.622</td>
<td>0.614</td>
</tr>
</tbody>
</table>

Significant correlations are shown in bold type; summary scales of each questionnaire in gray

Notes: The SRI-N domains were respiratory complaints (RC), physical functioning (PF), attendant symptoms and sleep (AS), social relationships (SR), anxiety (AX), psychosocial well-being (WB), social functioning (SF), and summary scale (SS). The SF-36 domain were physical functioning (PF), role physical (RP), body pain (BP), general health (GH), vitality (VT), social functioning (SF), role emotional (RE), mental health (MH), physical health component (PHC), and mental health component (MHC).
Criterion validity for the non-invasive HMV users

The analyses were also applied with the tracheotomised patients excluded. The correlation analysis also for this group reviled a significant correlation. The highest correlation was between physical functioning (PF) in both SRI-N and SF-36v2 and between SRI social functioning (SF) and SF-36v2 social function (SF).

Table 8. The correlation matrix for SRI and SF-36 v2 for the non-invasive HMV users.

<table>
<thead>
<tr>
<th></th>
<th>SF-36</th>
</tr>
</thead>
<tbody>
<tr>
<td>SRI</td>
<td>PF  RP  BP  GH  VT  SF  RE  MH  PHC  MHC</td>
</tr>
<tr>
<td>RC</td>
<td>0.437 0.551 0.345 0.614 0.495 0.503 0.346 0.263 0.557 0.341</td>
</tr>
<tr>
<td>PF</td>
<td>0.713 0.623 0.337 0.628 0.434 0.473 0.435 0.358 0.637 0.360</td>
</tr>
<tr>
<td>AS</td>
<td>0.258 0.266 0.503 0.381 0.404 0.308 0.212 0.276 0.419 0.214</td>
</tr>
<tr>
<td>SR</td>
<td>0.310 0.449 0.474 0.500 0.582 0.687 0.405 0.543 0.461 0.587</td>
</tr>
<tr>
<td>AX</td>
<td>0.356 0.450 0.425 0.494 0.399 0.613 0.398 0.512 0.427 0.521</td>
</tr>
<tr>
<td>WB</td>
<td>0.383 0.563 0.499 0.627 0.689 0.716 0.558 0.637 0.455 0.700</td>
</tr>
<tr>
<td>SF</td>
<td>0.438 0.633 0.509 0.599 0.557 0.705 0.501 0.451 0.580 0.569</td>
</tr>
<tr>
<td>SS</td>
<td>0.503 0.648 0.576 0.712 0.626 0.755 0.524 0.551 0.632 0.599</td>
</tr>
</tbody>
</table>

Notes: The SRI domain were respiratory complaints (RC), physical functioning (PF), attendant symptoms and sleep (AS), social relationships (SR), anxiety (AX), psychosocial well-being (WB), social functioning (SF), and summary scale (SS). The SF-36 domain were physical functioning (PF), role physical (RP), body pain (BP), general health (GH), vitality (VT), social functioning (SF), role emotional (RE), mental health (MH), physical health component (PHC), and mental health component (MHC).

Significant correlations are shown in bold type; in light gray, outstanding results for each scale, excluding summary scales; in dark gray, summary scales of each questionnaire.
Criterion validity for the patients receiving ventilation via a tracheotomoy

The tracheotomised patients in this sample only consist of 11 participants. Also for this group the correlation analysis reviled a significant between SRI-N sum score and SF-36 vitality (VT) \(r=0.874\) and between SRI –N sum score and the both sum scores of physical health complain PHC \(r=0.881\) and SF-36 mental health sum score MHC \(r=0.721\). These results demonstrate a substantial criterion validity for the SRI-N.

Table 9. The correlation matrix for SRI –N domains and SF-36v2 domains for the patient receiving ventilation via a tracheotomoy based on only 11 patients

<table>
<thead>
<tr>
<th>SF-36</th>
<th>SRI PF</th>
<th>RP</th>
<th>BP</th>
<th>GH</th>
<th>VT</th>
<th>SF</th>
<th>RE</th>
<th>MH</th>
<th>PHC</th>
<th>MHC</th>
</tr>
</thead>
<tbody>
<tr>
<td>RC</td>
<td>-0.174</td>
<td>0.412</td>
<td>0.393</td>
<td>0.668</td>
<td>0.834</td>
<td>0.579</td>
<td>0.735</td>
<td>-0.047</td>
<td>0.030</td>
<td>0.768</td>
</tr>
<tr>
<td>PF</td>
<td>0.428</td>
<td>0.527</td>
<td>0.203</td>
<td>0.560</td>
<td>0.487</td>
<td>0.203</td>
<td>0.538</td>
<td>-0.047</td>
<td>0.294</td>
<td>0.275</td>
</tr>
<tr>
<td>AS</td>
<td>0.000</td>
<td>0.000</td>
<td>0.281</td>
<td>-1.46</td>
<td>0.524</td>
<td>0.284</td>
<td>0.595</td>
<td>0.359</td>
<td>-0.566</td>
<td>0.585</td>
</tr>
<tr>
<td>SR</td>
<td>0.238</td>
<td>0.774</td>
<td>0.156</td>
<td>0.471</td>
<td>0.208</td>
<td>0.518</td>
<td>0.324</td>
<td>0.328</td>
<td>0.426</td>
<td></td>
</tr>
<tr>
<td>AX</td>
<td>-0.121</td>
<td>0.067</td>
<td>0.551</td>
<td>0.575</td>
<td>0.590</td>
<td>0.143</td>
<td>0.703</td>
<td>0.350</td>
<td>0.152</td>
<td>0.648</td>
</tr>
<tr>
<td>WB</td>
<td>-0.026</td>
<td>0.162</td>
<td>0.724</td>
<td>0.369</td>
<td>0.892</td>
<td>0.359</td>
<td>0.929</td>
<td>0.556</td>
<td>0.006</td>
<td>0.830</td>
</tr>
<tr>
<td>SF</td>
<td>0.373</td>
<td>0.585</td>
<td>0.112</td>
<td>0.511</td>
<td>0.290</td>
<td>0.224</td>
<td>0.086</td>
<td>-0.364</td>
<td>0.881</td>
<td>-0.267</td>
</tr>
<tr>
<td>SS</td>
<td>-0.104</td>
<td>0.324</td>
<td>0.590</td>
<td>0.579</td>
<td>0.874</td>
<td>0.322</td>
<td>0.841</td>
<td>0.377</td>
<td>0.881</td>
<td>0.721</td>
</tr>
</tbody>
</table>

Significant correlations are shown in bold type; in light gray, outstanding results for each scale, excluding summary scales; in dark gray, summary scales of each questionnaire.

Notes: The SRI domain were respiratory complaints (RC), physical functioning (PF), attendant symptoms and sleep (AS), social relationships (SR), anxiety (AX), psychosocial well-being (WB), social functioning (SF), and summary scale (SS). The SF-36 domain were physical functioning (PF), role physical (RP), body pain (BP), general health (GH), vitality (VT), social functioning (SF), role emotional (RE), mental health (MH), physical health component (PHC), and mental health component (MHC).
8.2.3.4. Construct validity

Most of the hypotheses were fulfilled.

**Hypothesis 1: SRI-N can differentiate between groups of patients with different diagnoses**

The SRI-N discriminated significantly between groups of patients as showed in table 10. There was a significant difference in the total sum score of quality of life (SRI.SS) between the four main diagnosis groups. A Krustal-Wallis test revealed a statistically significant difference between the four groups NMD patients, n=40, COPD patients, n=24, OHS patients, n=36, and the group of others patients, n=19 in the SRI.RC, SRI.PF, SRI.AX, SRI.WB and SRI.SS. A Bonferroni test were used for correction.

**Table 10. Differentiates in SRI domain between the four groups of patients**

<table>
<thead>
<tr>
<th></th>
<th>SRLSS</th>
<th>SRLRC</th>
<th>SRLPF</th>
<th>SRLAS</th>
<th>SRLSR</th>
<th>SRLAX</th>
<th>SRLWB</th>
<th>SRLSF</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chi-Square</td>
<td>14.41</td>
<td>17.79</td>
<td>9.87</td>
<td>3.22</td>
<td>7.19</td>
<td>14.62</td>
<td>10.52</td>
<td>13.38</td>
</tr>
<tr>
<td>Asymp. Sig.</td>
<td>.001</td>
<td>.000</td>
<td>.000</td>
<td>.200</td>
<td>.027</td>
<td>.001</td>
<td>.005</td>
<td>.001</td>
</tr>
<tr>
<td></td>
<td>.006 (a)</td>
<td>.000(a)</td>
<td>.006(a)</td>
<td>.202(a)</td>
<td>.026(a)</td>
<td>.001(a)</td>
<td>.005(a)</td>
<td>.001</td>
</tr>
</tbody>
</table>

a. Based on 100 000 sampled tables
Grouping Variable: HDIAGfiregr : NMD, COPD, OHS and others

**Hypothesis 2: COPD patients have more respiratory complaints than other groups**

A Mann-Whitney Test revealed a significant difference in respiratory complaints (SRI-N, RC) between COPD patients (Median=35, n=24) and OHS patients (Median = 62.5, n=37) \(z=-4.060, \ p=0.000\). Also between COPD patients (Median=35, n=24) and NMD patients (Median= 59.4, n=43)\(z=-3.398, \ p=0.001\) and between COPD patients (Median =35, n=24) and the group “Others” \( (Median \ 56.4, \ n=19)\(Z=-3.664, \ p<0,000\).
Hypothesis 3: COPD patients have more anxiety and depression than patients without COPD

A Mann-Whitney Test revealed a significant difference in anxiety (SRI-N AX) between COPD patients (Median=40, n=24) and OHS patients (Median=65, n=37) (z=-3.3327, p=0.001). Also between COPD patients (Median=40, n=24) and NMD patients (Median=70, n=43) (z=-3.525, p<0.000).

Hypothesis 4: Neuromuscular patients have poorer physical function than other patients

A Mann-Whitney Test revealed a significant difference in physical function (SRI-N.PF) between NMD patients (Median=29.2, n=43) and OHS patients (Median=50, n=37) (z=-2.601, p=0.009). Also between NMD patients (Median=29.2, n=43) and the group “others” patients (Median=45.8, n=19) (z=-2.343, p=0.019). But there was no significant difference between NMD and COPD in SRI.PF (z=-0.630, p=0.529).

Hypothesis 5: Patients receiving most hours of HMV per day have a worse HRQL

A) The relationship between hours of HMV per day and total score of quality of life was investigated using Spearman correlation coefficient. There was a significant negative correlation between the two variables r= -0.195, n=121, p=0.032.

B) The relationship between hours of HMV per day and physical function (PF) was investigated using Spearman correlation coefficient. There was a significant negative correlation between the two variables r= -0.358, n=125, p<0.000.

C) The relationship between hours of HMV a day and social function (SF) was investigated using Spearman correlation coefficient. There was a significant negative correlation between the two variables r= -0.196, n=121, p=0.029. In the others domains there were also a negative, but non significant correlations between the hours of ventilation and the domain of social function.
Hypothesis 6 Impairment of lung function has a negative influence on HRQoL

The relationship between impairment of lung function and HRQoL was investigated using both parametric and non parametric analysis. Both Spearman and Pearson correlation coefficient revealed significant correlation between VC and SRI-N.RC, SRI-N.PF, SRI-N AX and SRI-N sum score, and also between FEV1 and SRI-N.RC, SRI-N.PF, SRI-N AX and SRI-N sum score as showed in table. In addition Pearson correlation test showed a significant correlation between VC and SRI-N .SR and between FEV1 and SRI-N .SR

Table 11. Non parametric correlation
Spearman correlation coefficient

<table>
<thead>
<tr>
<th>Correlation between FEV1 and SRI</th>
<th>N</th>
<th>r</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>SRI.RC</td>
<td>89</td>
<td>0.338**</td>
<td>0.000</td>
</tr>
<tr>
<td>SRI.PF</td>
<td>89</td>
<td>0.356**</td>
<td>0.001</td>
</tr>
<tr>
<td>SRI.AS</td>
<td>89</td>
<td>0.007</td>
<td>0.949</td>
</tr>
<tr>
<td>SRI.SR</td>
<td>89</td>
<td>0.160</td>
<td>0.136</td>
</tr>
<tr>
<td>SRI.AX</td>
<td>89</td>
<td>0.291**</td>
<td>0.006</td>
</tr>
<tr>
<td>SRI.WB</td>
<td>89</td>
<td>0.161</td>
<td>0.138</td>
</tr>
<tr>
<td>SRI.SF</td>
<td>89</td>
<td>0.184</td>
<td>0.085</td>
</tr>
<tr>
<td>SRI.SS</td>
<td>89</td>
<td>0.271*</td>
<td>0.012</td>
</tr>
</tbody>
</table>

** Correlation is significant at the 0.01 level (2-tailed)
* Correlation is significant at the 0.05 level (2-tailed)

Significant correlations are shown in bold type

Table 12. Non parametric correlation
Spearman correlation coefficient

<table>
<thead>
<tr>
<th>Correlation between FC and SRI</th>
<th>N</th>
<th>r</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>SRI.RC</td>
<td>89</td>
<td>0.276**</td>
<td>0.010</td>
</tr>
<tr>
<td>SRI.PF</td>
<td>89</td>
<td>0.33**</td>
<td>0.001</td>
</tr>
<tr>
<td>SRI.AS</td>
<td>89</td>
<td>0.039</td>
<td>0.718</td>
</tr>
<tr>
<td>SRI.SR</td>
<td>89</td>
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<td>0.019</td>
</tr>
<tr>
<td>SRI.AX</td>
<td>89</td>
<td>0.318**</td>
<td>0.003</td>
</tr>
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</tr>
<tr>
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<tr>
<td>SRI.SS</td>
<td>89</td>
<td>0.296**</td>
<td>0.006</td>
</tr>
</tbody>
</table>

** Correlation is significant at the 0.01 level (2-tailed)
* Correlation is significant at the 0.05 level (2-tailed)
9.0 Discussion

The major finding of the present study was that the Norwegian version of SRI, the SRI-N, is a reliable and valid questionnaire for patients receiving HMV in NIV mode. In addition there are indications that SRI-N also seem to be valid in assessing quality of life for patients receiving HMV via tracheostoma. However the small number of these patients and the specific problems in this group, leads to a necessary reservation concerning the conclusions of the results from this group of patients.

Translation process

In the process of translationing the instrument into Norwegian, general guidelines for transcultural adaptations of quality of life instruments were followed (Guillemin, Bombardier, & Beaton, 1993; Herdman, Fox-Rushby, & Badia, 1997, 1998; Kvamme, 1998; Wahl & Hanestad, 2004).

The translators had different backgrounds, but they were all familiar with the goal of the questionnaire. Three of them were licensed translators; one of them had German as first language. The physician who translated it into Norwegian had extensive knowledge of HMV. He also had German as first language. The differences in background may have given a well formulated translation. A translation/ back translation procedure was followed. When translating back to German, the word “bound” in item 15 was changed to “tied”. There is a difference in “being bounded” to your home and “being tied” to your home. This could lead to a misinterpretation of this item. Item 15 is one of eight items in the Social functioning scale and internal reliability of this scale was therefore assessed both with and without this item. With item 15 the Chronbach alfa was 0.79 without it the Chronbach alfa was 0.80. This can indicate that the responders had interpreted the questionnaire in the way it was meant. Translating the German word “Luftnot” was also a challenge. It was translated into “åndenød” and “tungpusthet”, meaning “shortness of breath” and “difficult breathing”. The semantically correct translation would be “åndenød”, but this is a somewhat old-fashioned word, which is not so common in today’s everyday spoken Norwegian. Since there were some disagreements about which term to use, it was first concluded that both terms should be included in the Norwegian translation. After a discussion it was decided that it was best to strive to achieve term-wise equivalence. Therefore the Norwegian “tungpusten” would cover the meaning of the German “Luftnot” (shortage of breath).
The challenge in translating a questionnaire is to find words and expressions which exactly cover the original text. The rules of translation include syntax, semantics and pragmatics conditions. Syntax refers to the rules of how words are put together in a sentence. The semantics examination tries to determine the meaning of the words (or how they are put together). While a pragmatic research looks on the communication in its context. The pragmatic approach concludes that syntax and semantic rules are not sufficient to determine the meaning of words. Isolated words and expressions have only a latent meaning. The words obtain active meaning when they are included into a context (Munitz, 1981).

In this study accepted procedures for translations and cultural adaptations of QoL questionnaire were followed. To ensure good quality of the trans-cultural adaptation, both professional health care workers with long time experience in HMV and patients were asked to see if the questionnaire was clear and easy to understand and that it was relevant and covered topic of interest.

In the pilot study the patient group was represented by the Norwegian organization for HMV users (Respira). The group is representative in relation to sex, age and diagnosis, but as members of a patient organization they may have a larger awareness and consciousness in relation to empowerment and co-determination. Asking the group that represents the HMV users may present bias but at the same time their participants was very helpful as they have a genuine interested and want to influence on issues that concern this group. They also had wise and useful remarks to the SRI-N questionnaire.

The intention of including the patient organization was also to meet some of the criticisms in QoL research claiming that the target groups situation in life, problems and challenges are not mapped out well enough. (Bowling, 2001). It is therefore important that the items in SRI-N make sense for the members in Respira. They reported that the questionnaire was easy to understand and that it could bee self-administered. However one of the responders had some comments on the questionnaire. As a wheelchair user he found the item “is it difficult for you to use the stairs “difficult to answer. In his opinion the answer alternative” do not fit” was not precise. He claimed that “impossible” also should have been an alternative. He also maintained that the questionnaire did not sufficiently cover the fact that a large number of patients receiving HMV are considerably disabled. His experience was that the SRI questionnaire focused to much on
and was addressed to HMV users breathing problems but not all the other factors that impact the wheelchair users daily living.

The Respira group also wanted to investigate whether education in using the HMV equipment and the service and follow up from the Public Health Services were satisfactory. Therefore questions concerning these issues were included in the specific questionnaire. Satisfaction with care is an important and often neglected component in disease specific and generic scales or battery of scales. A persons degree of satisfaction with their health status and outcome and fulfilment of expectations of the treatment should be included (Bowling, 2001).

It is important to listen to comments and objections to the questionnaire from HMV users. But the advantages of having an international equivalent questionnaire would not be maintained if the meaning or the answering alternatives in SRI-N were changed.

The translation of a HRQoL questionnaire is not a simple operation as it is subject to one overriding requirement -equivalence between the source and target version. It also subjected to constraints of cost and time. It must be remembered that the translation conditions the result of future research. If items are poorly worded or not understood correctly, the execution of the study and the interpretation of results will be adversely affected and distorted (Acquadro, 2004).

The aim of the translation was to make an identical version of the original SRI but adapted to Norwegian conditions. SRI-N has a simple language; the expressions are short and concrete and are in an active form. The adaptation that was done was based upon everyday speech in Norway and Germany.

Despite the comment from one of the members in the Respira the SRI-N was experienced as understandable and clear both from the professionals and the target group.

**Psychometric properties of the SRI-N**

The psychometric testing of SRI-N can be seen as an evaluation of the translation and cultural adaptation and the new instruments performance. The different steps in the validation process will be discussed.

**Administration**

In the present study the questionnaires was sent by mail to those in the Norwegian registry of HMV users who met the inclusion criteria. In the German and Spanish validation studies the patients were recruited to the studies during a routine visit at their outpatient clinic. They also completed the questionnaire in the presence of the researcher, who had the opportunity to
intervene if the patient had any questions or doubt about being able to complete the questionnaire.

The administration of a questionnaire may affect the technical equivalence (Wahl & Hanestad, 2004). It may be an advantage to meet the patient and be able to help filling out the questionnaire. However it is emphasized that SRI is self-administered questionnaire. In the present study the participant were informed that they could contact the researcher if they had any questions or needed help answering the questionnaire.

The differences in the way of administration the Norwegian and the Spanish/German version of the SRI, may affected the technical equivalence in SRI-N which imply that the way of collecting data may have an impact on the result.

However it may seem that bias effect caused by an other phenomena “Social Desirability Responding” (tendency to idealize your life) may be less in force when the questionnaire is self-administered compared with administration mode that imply meeting between the researcher and the participants (Næss, 2001). Mailed self-administered questionnaire, is an economical way of collecting data but not appropriate for surveying certain populations e.g., children and elderly (Polit & Beck, 2004). The HMV patients are followed with medical control in the outpatient clinic between every third months and every year. This study needed to be completed within a limitation of time and recourses therefore the questionnaire was mailed to the sample.

**Participants and non-participants**

There were no significantly differences between participants and non-participants regarding age, sex, different diagnosis and year of HMV. However there was a significant difference in FVC and FEV1 between responders and non responders. The analysis showed that non-participants had a significant lower FVC and FEV1 than the participants. It may be that the non responders did not answer the questionnaire because they had a more severe disease and their health conditions were worse than the responders. When correlating the lung function FEV1 and FC with SRI-N SS it revealed a significant correlation between the lung function and SRI sum score, suggesting that the non-responders also have lower HRQoL than the participants. This result may be interesting because earlier studies have shown inconsistent degrees of correlation between physiologic parameters e.g. FVC and FEV1, and QoL (Yessen, 2000).
Sample

The sample population included patients with the most common diagnosis for starting HMV. In the earlier validation studies of SRI questionnaire tracheotomised patients were not included (Lopez-Campos et al., 2008; Windisch et al., 2003b). However the authors of these studies suggest it would be of interest to study HRQL in tracheotomised patients compared with non invasive ventilated patients to see if the SRI can discriminate between these two groups.

A direct comparison of QoL between NIV and tracheotomised IPPV users is difficult. There have been no randomized trial and there are not likely to be any as patients who are treated with invasiv ventilation are likely to have more advanced disease, greater ventilator dependency and often bulbar disease (Simonds, 2007).

In the present study it was chosen to validate the SRI including both non-invasive and tracheotomised patients. The tracheotomised patients might have been excluded, but as far as we know there is no specific questionnaire developed for tracheotomised patients receiving HMV. Only a specific instrument for children and their caregivers is developed and validated (Hartnick et al., 2002).

The tracheotomised patients are a small but important group in the population of HMV patients in Norway. Many of the ethical challenges are related to this group. It would be useful and important to get some knowledge about whether the SRI is suitable for this group. Although experience and evidence indicate that NIV have advantages over non-invasive ventilation (Bach, 1995; Bach, 2004) Markstrom found that patient ventilated via a tracheotomy with skoliose, post polio respiratory failure and other neuromuscular disease, had higher QoL score than in the NIV group (Markstrom et al., 2002).

Reliability

To test reliability, the internal consistency was analyzed. The internal consistency is an item correlation test that shows the homogeneity of the items. Cronbach alpha coefficient above 0.7 is generally regarded as acceptable for psychometric scales (Fayers & Machin, 2007).

Cronbach alpha coefficient obtained for the total sample in each domain varied from 0.761 to 0.884. Because of possibility of misinterpretation of item 15, chronbach alfa also was applied in SF domain without item 15. The punctuation was then 0.80. Cronbach alpha coefficient
obtained for the sample without the tracheotomised patients varied from 0.66 to 0, and 89. In the Spanish translation of SRI the Cronbach alpha coefficient was >0.7 for all scales except social relationships (Lopez-Campos et al., 2008). This indicate an acceptable or homogeneity between the items in the Norwegian version of SRI. However it is important to be aware that an instrument can be reliable and homogeneous regardless of whether it is suitable for assessing what it was meant to measure.

Validity
The result of the study shows that the criteria for content validity, criterion validity and construct validity are fulfilled.

Content validity
The content validity in the present study is based on following the rigorous procedures for translation and back translation. To ensure face validity both professional health care workers with long experience in the field and members of the target group took part in the process of producing the SRI-N. The target group was represented by members of the patient organization Respira.

Earlier studies have shown that there has been no agreement over what relevant QoL domain that should be measured in respiratory diseases, and that the criteria for assessment vary between studies. The lack of consensus is described as partly due to the lack of qualitative descriptive data of the lives of these patients. Without such data the relevance of existing scales remains problematic and not assessable (Bowling, 2001). Concerning SRI these challenges were met by the process of developing the original SRI, with a combination of a panel of experts who ensured the professional content and methodology and patients who were interviewed to get the necessary description of their lives (Windisch et al., 2003b).

The need for validation tests has to be considered separately for the particular study through the research question, the method and the sample (Kvamme, 1998).

The strategy for validation in the present study is based on the validation tests used in the validation study of the original SRI questionnaire and in the Spanish validation study (Lopez-Campos et al., 2008; Windisch et al., 2003b).
Criterion validity

Criterion validity involves assessing an instrument against the true value (Fayers & Machin, 2007a; Streiner & Norman, 2003) The “true” value in quality of life measurements is not easy to define. Studies shows that observers misjudge both symptoms and general QoL. Health care workers tend to base their observations of overall QoL upon physical signs such as symptoms, and both over- and underestimating the patients’ condition occurs (Fayers & Machin, 2007b).

Concurrent validity means agreement with the true value. SF-36 was chosen as standard because it was used both in developing SRI (Windisch et al., 2003b) and in the Spanish translation (Lopez-Campos et al., 2008). Its psychometric properties are well documented (J. E. Ware et al., 2008). SF-36 is translated and validated to Norwegian, by Loge et al (Loge & Kaasa, 1998).

As expected a significant correlation was confirmed by the correlation analysis between scales of the SRI and scales of the SF-36 for the totally sample. Spearman rank correlation revealed the highest correlation between physical functioning SRI (PF) and SF-36v2 (PF) \( r=0.729 \) and between SRI Well-being (WB) and SF-36v2 vitality (VT) \( r=0.713 \), also between the most of the other domains there were a high correlation. The large correlation \( r=0.729 \) in SRI PF and SF-36 PF in both the total group and in the non-invasive group is important because it can be assumed that SRI measure aspects of physical functioning which is an important domain and are related to activities of daily living.

In the first validation study of SRI the correlation of subscales of the SRI and the SF-36 was \( 0.21<r<0.79 \). The highest correlation was found between SRI-Psychological Well-Being and Mental Health of the SF-36 \( r = 0.79 \). The lowest correlation was found between SRI-Attendant Symptoms and Sleep and Role-Emotional of the SF-36 \( r = 0.21 \). (Windisch et al, 2003b). Exact the same correlation between SRI (AS) and SF-36 (RE) was found in the non invasive group of patients in the present study.

The sample in the present study consists of both non-invasive and tracheotomised patients. Correlations test were done regarding the total sample and also for each of the two groups. For tracheotomised there was a medium correlation \( r=0.428 \) between SRI .PF and SF-36 PF. The tracheotomised have NMD and particular conditions with their disease. The most interesting result concerning this group is the large correlation between SRI sum score and the two sum scores of SF-36 physical health complain PHC \( r= 0.881 \) and mental health
sum score MHC (\( r=0.721 \)). The interpretation of this result may indicate that SRI can be used as an instrument for these patients, but the group is too small to generalize it to the total population. Studies with a larger sample are needed.

There was also a large correlation in both the non-invasiv and the tracheotomised patients in SRI well being (WB) and SF-36 vitality (VT). These aspects are important because they affect patients' lives in many ways. Siri Næss emphases in her definition of QoL the subjective experience to having good feelings and positive evaluations of ones own life. Good feelings can be joy, energy and love being satisfied, having self-respect and experience a meaning of your life (Næss, Moum, Mastekaasa, & Sørensen, 2001). These issues are especially important in a disease specific questionnaire that often trend to focus on negative effects of a disease or a condition.

**Construct validity**

The construct validity is one of the most important characteristics of a measuring instrument. It is an assessment of the degree to which an instrument measure the construct it was meant to measure. The construct validity is measured trough fulfilling all the established hypotheses (Fayers & Machin, 2007a). It was hypothesised that the SRI-N was capable of discriminate between diagnosis and group of patients. The results demonstrated that the SRI was able of discriminating between the four main groups of patients. The HRQoL is strongly influenced by underlying disease. This is consistent with the findings of the German and the Spanish validation study. Interpretation of this result may lead to the assumption that SRI-N can be seen as both a condition specific and a disease specific questionnaire. It is related both to the condition of being a HMV user and it is able to discriminate between the characteristic that are related to the specific disease.

The hypothesis was that COPD patients have more respiratory complaints than other groups. It was expected because COPD patients who require HMV suffer from chronic respiratory failure and from additional lung disease. The results demonstrated that the patients with COPD have more respiratory complaint than other patients. These findings are also supported by those of Voll-Anerud who examined the relationship between QoL and number of respiratory symptoms and impaired levels of both the PCS and MCS (Voll-Aanerud, Eagan, Wentzel-Larsen, Gulsvik, & Bakke, 2007, 2008).
The hypothesis that COPD patients have more anxiety and depression than patients without COPD was also fulfilled. These findings are consistent with those of Meechan who found higher levels of depression and anxiety in the COPD group than other groups (Meechan, 1995).

The hypothesis that numbers of hours of ventilation had an impact on QoL was also confirmed by the result. These results are consistent with the findings in previous studies with patient receiving HMV via non-invasive ventilation. (Lopez-Campos et al., 2008; Windisch et al., 2003b).

The last hypothesis was that impairment of lung function has a negative influence on QoL. Earlier studies have shown inconsistent results of correlation between physiologic parameters e.g. FVC and FEV1 and QoL (Yessen, 2000). One study examined if it was possible to localise the benefit of an intervention by analysing the various domain in a questionnaire. The SF-36 score was not as sensitive as SGRQ in detecting these changes and the improvement in FEV1 was small and only weakly correlated with changes in SGRQ score. These findings are also supported by those of Miravitlles who found that having FEV1 lower than 50% predicted was related to lower HRQoL measured by the St. George’s Respiratory Questionnaire (SGRQ) (Miravitlles et al., 2005). To our knowledge the present study is one of few that find this relationship between QoL and lung function.

Only four patient receiving ventilation via tracheotomy had recent measured VC and FEV1, hypothesis number 6 was therefore not tested for patients with tracheostomy.

The results show that all the six hypothesis were fulfilled, this suggests acceptable construct validity for SRI-N.

In the first validation study of SRI construct validity was confirmed by factor analysis, indicating one summary scale that accounts for 59.8% of the variance (Windisch et al., 2003b). In the present study Explorative factor analysis for SRI, principal axis factoring, varimax-rotasjon: explained 67 % of the variation and revealed 13 factors which is the same as revealed in the Spanish validation study (Lopez-Campos et al., 2008). When factor analysis were applied with 7 factors the result explained 55% of the variation of the questionnaire.
Factor analysis of the questionnaire, resulted in 13. If there should be different divisions in the different domains between in the German, Norwegian and the Spanish version of the SRI, it would not be possible to compare the result between the different countries. Therefore the intention of doing the factor analysis was to confirm the German version, but for this purpose the study group was too small.

**Responsiveness**

Responsiveness of SRI was not measured in the present study because it is related to changes within patients and demands a prospective or a longitudinal study design. However a highly sensitive scale will usually also be highly responsive (Fayers & Machin, 2007a). Recent published results shows that the original SRI revealed a high responsiveness for changes in HRQoL after initiating HMV (Windisch, 2008). These results are important because there have been conflicting results concerning the effect on HMV especially in COPD patients. This may be because the previous studies were used instruments which were not developed for measuring HRQoL patients with severe respiratory insufficiency receiving HMV. Also The Norwegian National Council for Quality and Prioritising in the health services has claimed that the treatment is not beneficial for COPD (Nasjonaltråd & helsetjenesten, 2008). The new findings indicate that HMV have a positive effect on HRQoL also in patients with COPD (Windisch, 2008).

**Source of bias with translation of SRI**

There are several general bias related to using questionnaire in QoL research. The bias can be connected both to the persons that answer the questionnaire and to the questionnaire itself. Bias can be caused by Social Desirability Responding with is a tendency to idealize your life (Næss, Moum, Mastekaasa, & Sørensen, 2001). Selective reporting can present a bias problem when using normative data. Tendency of resistance to extreme scores and to answer based on cultural expectations and acceptable norms can occur. Change in self-evaluation in patients with a considerably degree of disease also can be seen as bias (Wahl & Hanestad, 2004). Although response shift is less important if perceptions are considered to matter more than objective disease. In QoL research it is rarely possible to quantify the overall bias, and these effects cannot be separated (Fayers & Machin, 2007).
Unintelligible meaningless or too many items may also cause bias (Wahl & Hanestad, 2004). It also seems that the respondents answer questionnaires even if they do not understand the item (Mallison, 2002). The tendency is also to answer in a homogeny way so that lack, of understanding may be difficult to detect by testing Internal consistency (Mallison, 2002).

Self-administration also makes it impossible to ask direct questions. Even if the respondents were informed that they could contact the persons behind the study, none of the respondents asked about the meaning of some of the items.

Item missing is a good indication of ambiguity and vagueness in the questionnaire (Fayers & Machin, 2007). Analysis of pattern of missing QoL items suggest that they often occur at random. In contrast intentional omission can occur if the item present particular difficulties for example item about “sexual interest” or any sensitive issue. Therefore it is likely to be more reticent concerning this kind of item. There are no items about sexual interest in SRI but there my bee item of sensitive character like “are you feeling lonely”. Other reason for missing may be that none of the answer alternative makes sense or that there are too many items (Fayers & Machin, 2007).

78 of 127 responders have completed the questionnaire without any missing items. Only 79 of the responders have answered item 31 (“My partner suffers because of my disease”). When considering that only 70 of the responders answered that they were married or live-in relationship, it may be possible that omission of this item is because the responders do not have any partner. Some of these missing can be explained because the responders might have been unsure regarding what answer alternative to chose. Nevertheless this issue has a marginal importance for the total result.

Differential item functioning (DIF) analysis could be used to test whether translations of items in multi-item scales are equivalent to the original, but DIF analysis is still rarely used for this purpose in health status assessment (Petersen et al., 2003). DIF analysis has not been used in the present translation process in the present study. It may have been an advantageous to have used this method as a reassurance in the translation of SRI.
Implications for clinical practice

Evidence based practice is using the best existing knowledge and putting it into practice with the best clinical experience and critical thinking skills to individualizing research results to best meet the needs of the patients (Bydam, 2003). Obtaining integration between research and evidence based practice, appropriate tools are needed. The SRI –N is a validated instrument that can be used to get new knowledge in QoL in patient with respiratory failure. Whether or not the process of validation was useful will depend on how the SRI-N is used in the future.

HMV are a very heterogenic group. Some of HMV users have diseases that do not influence daily life, whereas other HMV users suffer from end-stage disease. Assessment of subjectively reported HRQL is extremely important among the severely sick patients but also in those with fewer symptoms from their CRF. Having a Norwegian version of the SRI instrument makes it possible to obtain sufficient outcome for these patients. It also makes it possible to evaluate if the main goal for the Norwegian National centre of home mechanical ventilation. That is to maintain or increase the HRQoL for HMV patients in Norway.

Critical remarks

In the present study the questionnaire was sent by mail to those in the Norwegian Registry of HMV users who meet the inclusion criteria. The significant difference in FEV1 between responders and non responders shows that it may have been better to include these patients when they attended the outpatient clinic. The difference between the responders and no responders may also be a sign of how severely ill many of these patients are and that is demand much effort to answer a questionnaire.

There are studies that conclude that it should be developed disease-specific QOL measure for ALS patient receiving HMV which does not focus solely on health status but more generally assessment of QOL in a palliative care situation. When patient are cared for at home also the primary caregivers should be included in order to get the full picture (Kaub-Wittemer, Steinbuechel, Wasner, Laier-Groeneveld, & Borasio, 2003). These moments are important to have in mind, still there will be an advantage of using a common condition specific questionnaire for all HMV users. E.g. is it interesting to discover findings that indicate that
ALS patient had a better sum score than other NMD patient. This may be explained by the influence of the response shift, and illustrates that there are other factors than health and severe disease that have influence and impact on QoL (Fayers & Machin, 2007a; Richard & Folkman, 2000; Schwartz & Sprangers, 2000; Wahl & Hanestad, 2004).

**Conclusion**

Based on the result of this study the Norwegian version of the SRI, the SRI-N questionnaire is a reliable and valid condition specific questionnaire for use in the Norwegian HMV population of patients with NIV. Translation and cross-cultural adaptation and the psychometric properties of this instrument allows for its application in clinical practice and research within Norway as well as for comparative international studies. For tracheotomised patients the questionnaire may be valid, many of the validation criteria were fulfilled. However, as there were only 11 tracheotomised responders further research in the total sample of tracheotomised patients in Norway is needed.

**Future Aspects**

A validated instrument such as the Norwegian version of the SRI gives a very interesting opportunity to conduct a prospective or longitudinally study design to assess changes in HRQoL after establishing HMV.

It will be possible to assess HRQoL in the total group of Norwegian HMV patients. This would be an important part of the evaluation of the efficacy of this treatment and the first time such a study was performed with a disease specific questionnaire in Norway. This may provide new knowledge in HRQoL outcome in HMV treatment that is important also in terms of health policies addressing some of the questions. The Norwegian national council for quality and prioritising in the health services have asked previously (Nasjonaltråd & helsetjenesten, 2008). It will also be important to assess if living at home or in an institution affect the perceived HRQoL.

In addition to SRI-N and SF-36 v2 the participants in the present study also answered a questionnaire about how they experienced the follow up from both the primary and secondary
health care services. These data could serve as important additional markers of the quality of services provided by both primary and secondary healthcare levels and possibly identify areas which should be emphasized in future directives for treatment goals and indications.

The questions were related to how the HMV users experienced the education in use of the equipment and what kind of special problems they experience with the use of mask and the tracheotomy. Analysis of these data will provide new knowledge about how to organize the Health services can improve both education and follow up in a better way for these patients.

The Severe Respiratory Insufficiency (SRI) questionnaire was developed specially for HMV patients. The questionnaire contains items addressing problems that patients with CRF experience. The questionnaires were developed in a group of patients with CRF of different origin, already treated with HMV for a longer time period. Reliability and validity of the SRI has not been investigated in a homogeneous group of patients who actually suffer from CRF prior to treatment with HMV and who were not treated with HMV (Duiverman, Wempe, Bladder, Kerstjens, & Wijkstra, 2008). This suggest a possible role for the SRI in future research regarding HRQoL in patients with CRF without HMV.

The Respira group emphasized the importance of communicating results from the study to the HMV users. It is important to continue the cooperation with the patient organization in future research within this group. This issue is also emphasized by Fayers, to collect information in a form that can be communicated to future patients, thus enabling them to anticipate and understand the consequences of their illness and its treatment options (Fayers & Machin, 2007). This could contribute to the patient’s ability and desire to take specific actions and make decisions of their own (HOD, 2001a).
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Appendix

Appendix 1
Acceptance of the study from The Norwegian Data Inspectorate

Appendix 2
Acceptance of the study from The Regional Ethical Research Committee

Appendix 3
Acceptance of the study from The Norwegian Registry of patients receiving HMV (2006)

Appendix 4
Acceptance of the study from The Norwegian Registry of patients receiving HMV (2008)

Appendix 5
Information letter to the HMV patients

Appendix 6
The Norwegian version of The Severe Respiratory Insufficiency Questionnaire, SRI questionnaire

Appendix 7
Scoring Procedure for the SRI

Appendix 8
Demographic and specific questionnaire
Appendix 1

TIL RÅDING AV BEHANDLING AV PERSONOPPLYSNINGER

Vi sticker til melding om behandling av personopplysninger, motatt 27.12.2006. Meldingen gjelder prosjektet:

16001  

**16001**  

*Hoveden omfaner venke henners respiratoriske system, men også andre fagfrike fagområde onde fagområde. Oss analyse og regulering av rykk spørreskjema om livskvalitet har henners respiratoriske system.*

Behandlingsansvarlig: Universitetet i Bergen, ved institusjonene ansvarlige leder

Daglig ansvarlig: Berit Røkne Hansen

Student: Heidi Oknes Markussen

Personvernombudet har vurdert prosjektet, og finner at behandlingen av personopplysninger vil være regulert av § 7-27 i personopplysningsloven. Personvernombudet vil ikke at prosjektet gjenomføres.

Personvernombudets tillegg foreslår at prosjektet gjenomføres i tråd med opplysningene gitt i meldelser, korrespondanse med ombudet, eventuelle kommentarer samt personopplysningsloven/-s behandlingsloven med forskriften. Behandlingen av personopplysninger kan sees i gang.


Personvernombudet har lagt ut opplysninger om prosjektet i en offentlig database, http://www.ud.uib.no/personvern/register/


Vennlig hilsen

Bjørn Hesthagen

Siv Midthassel

Kontaktperson: Siv Midthassel tlf. 55 58 33 34

Veileg: Oppbygningstidning

Kopi: Heidi Oknes Markussen, Falkangervegen 11, 5108 HORDVIK
Ad. prosjekt: Hvordan opplever voksne hjemmerespiratorbrukere i Norge sin livskvalitet og hvilke faktorer påvirker deres livskvalitet. (273.06).

Det vises til din søknad om etisk vurdering datert 14.11.06. REK Vest vurderte studien i møte den 30.11.06.

Komiteen mener dette er en velfundert studie, men har følgende merknader.

I informasjonsskrivet bør ordet inviteres utgå.
Det mangler informasjon om kodeøkkel blir oppbevart. Hvis så er tilfelle, er dataene ikke anonyme men avidentifiserte. Dette må i så fall endres i informasjonsskrivet.
I samtykkeerklæringen bør avsnitt 2 og 3 utgå.

Med disse merknadene er studien endelig klarert fra denne komité sin side.

Vi ønsker dere lykke til med gjennomføringen og minner om at komiteen setter pris på en sluttrapport, eventuelt en kopi av trykt publikasjon når dette foreligger.

Med vennlig hilsen

Jon Lekven
leder

Marit Nedreli
sekretær
ERKLÆRING VEDRØRENDE BRUK AV PASIENTREGISTER

Som leder for nasjonalt kompetansesenter for hjemmerespiratorbehandling og ansvarlig for vårt pasientregister bekrefter jeg herved at vi vil samarbeide med Heidl Markussen i forhold til mastergradsoppgaven "Livskvalitet hos Hjemmerespiratorbrukere" og gi tillatelse til bruk av registeropplysninger etter godkjenning av prosjektet fra regional etisk komité.

Mvh

Ove Fondeves
Senterleder
Nasjonalt kompetansesenter for hjemmerespiratorbehandling
Vedr. søknad datert 27.6.2008 om å benytte opplysninger fra Nasjonalt register for hjemmerespiratorbehandling.

Vi har mottatt din søknad om tillatelse til å benytte opplysninger fra Nasjonalt register for hjemmerespiratorbehandling i masteroppgave ved Institutt for samfunnsmedisinske fag, Seksjon for sykepleievitenskap, Universitetet i Bergen.

Tema for oppgaven: Livskvalitet hos hjemmerespiratorpasienter.
Hovedveileder: professor dr. polit Brit Rokne Hønestad.
Medforfatter: statistiker Tore Wenzel-Larsen.

Prosjektet er godkjent av regional etisk komité (REK).

Opplysninger fra Nasjonalt register for hjemmerespiratorbehandling t.o.m. 31.12.2007 ble ferdig kvalitetsikkert 1.9.2008 og opplysningene er nå klargjort for analyser.

Din søknad er innvilget.

Du vil få tildelt en anonymisert SPSS fil med opplysninger fra registeret t.o.m. 31.12.2007 over hjemmerespiratorbrukere > 18 år i Hordaland, Rogaland og Sogn og Fjordane med følgende variabler:

<table>
<thead>
<tr>
<th>Nr.</th>
<th>Variabel</th>
<th>Labell</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>HDIAG</td>
<td>Hoveddiagnose</td>
</tr>
<tr>
<td>28</td>
<td>DATOOPPS</td>
<td>Dato for start av behandlingen (dato, måned, år)</td>
</tr>
</tbody>
</table>

**LUNGEFYSIOLOGISKE PARAMETER FOR BEHANDLINGSSTART**

<table>
<thead>
<tr>
<th>Nr.</th>
<th>Variabel</th>
<th>Labell</th>
</tr>
</thead>
<tbody>
<tr>
<td>30</td>
<td>FVK</td>
<td>FVK (liter)</td>
</tr>
<tr>
<td>31</td>
<td>FEVI</td>
<td>FEVI (liter)</td>
</tr>
<tr>
<td>34</td>
<td>PO2DAG</td>
<td>PO2 dagtid, arteriell (kPa)</td>
</tr>
<tr>
<td>35</td>
<td>PCO2DAG</td>
<td>PCO2 dagtid, arteriell (kPa)</td>
</tr>
<tr>
<td>36</td>
<td>PCO2NATT</td>
<td>PCO2 nattestid, arteriell (kPa)</td>
</tr>
<tr>
<td>37</td>
<td>HOYDE</td>
<td>Høyde (cm)</td>
</tr>
<tr>
<td>38</td>
<td>VEKT</td>
<td>Vekt (kg)</td>
</tr>
</tbody>
</table>
Det er ikke førende, men ønskelig fra registerets side at opplysninger fra registeret publiseres i nasjonale eller internasjonale vitenskapelige tidsskrifter, framfor i en monolog.

Lykke til med en spennende masteroppgave!

Vennlig hilsen

Elin Tollefesen
Overlege dr.med.
Forskningsansvarlig Nasjonalt register for hjemmerespiratorbehandling
Nasjonalt kompetanseenter for hjemmerespiratorbehandling og Lungeavdelingen
Haukeland Universitetssykehus

Vedlegg:
Dokument: Nasjonalt register for hjemmerespiratorbehandling; Retningslinjer for tildeling av analyserettigheter for data fra Nasjonalt register for hjemmerespiratorbehandling.
Forespørsel om å delta i undersøkelsen om hvordan det er å leve med hjemmerespirator eller annet hjelpemiddel til pustestøtte

Undersøkelsen du forespørs å delta i omhandler hvordan det er å leve med hjemmerespirator eller annet hjelpemiddel til pustestøtte som eksempel BIPAP. Bakgrunnen for at jeg henvender meg til deg er at du er registrert i Nasjonalt Kompetansesenter for Hjemmerespiratorbehandling (NKH) sitt register som bruker av et slikt hjelpemiddel.

Studiens hensikt

Helsevesenet i Norge mangler i dag opplysninger og kunnskap om hvordan det er å leve med hjemmerespirator eller annet hjelpemiddel til pustestøtte. For at vi skal gi best mulig tilbud til deg og andre patienter med denne type hjelpemiddel er det viktig med informasjon fra deg om hvordan det er å leve med hjemmerespirator eller annen form for hjelpemiddel til pustestøtte. Dette vil kunne danne grunnlag for kunnskapsbasert praksis hos helsepersonell. Hensikten med undersøkelsen er også å tilpasse et spørreskjema som er benyttet i flere europeiske land for norske forhold.

Gjennomføring av undersøkelsen og hva dette innebærer for deg


Taufshetsplikt

Frivillig deltakelse

Prosjektgruppe
Undertegnende er masterstudent ved Institutt for samfunnsmedisinske fag ved Universitetet i Bergen og arbeider også som sykepleier ved Haukeland Universitetssykehus og er tilknyttet Nasjonalt Kompetansesenter for hjemmerespiratorbehandling som prosjektmedarbeider. Professor Berit Rokne Hanestad ved Universitetet i Bergen er min hovedveileder. Leder av Nasjonalt Kompetansesenter for hjemmerespiratorbehandling og overlege Ove Fonedenes og dr.med Jon Hardie ved Haukeland Universitetssykehus er biveileder.

Har du spørsmål eller trenger hjelp til å fylle ut spørreskjemaet ta gjerne kontakt. Jeg kan treffes på telefon 55973549/ mobil 97499915 eller send e-post til heidi.markussen@helsebergen.no

Vennlig hilsen
Heidi Øksnes Markussen
Masterstudent /sykepleier

Vedlagt:
1. Spørreskjema
Følgende spørsmål berører den generelle helsetilstanden din. Utsagnene nedenfor tar for seg forskjellige aspekter ved det daglige liv.

Hvordan har du hatt det i løpet av *den siste uken*? Vennligst sett et kryss ved svaret som passer best med HVERT enkelt utsagn.

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt</th>
<th>Stemmer nesten ikke</th>
<th>Stemmer til en viss grad</th>
<th>Stemmer ganske bra</th>
<th>Stemmer helt</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Det er vanskelig å gå i trapper.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Jeg har tungpust under måltider.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Jeg føler meg ofte dårlig.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Selv uten fysiske anstrengelser har jeg pustevansker.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Jeg har ofte hodepine.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7. Jeg har mange venner og bekjente.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8. Jeg er bekymret for at sykdommen min skal bli verre.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10. Jeg kan godt omgås andre mennesker.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>11. Jeg er av og til svimmel.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>13. Jeg er redd for å få pustevansker om natten.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>14. Jeg har ofte vondt i nakken.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>15. Jeg er sterkt bundet til hjemmet mitt.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>16. Husarbeid er vanskelig for meg.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

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Hvordan har du hatt det i løpet av *den siste uken*? Vennligst sett et kryss ved svaret som passer best med HVERT enkelt utsagn.

<table>
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<th>Stemmer til en viss grad</th>
<th>Stemmer ganske bra</th>
<th>Stemmer helt</th>
</tr>
</thead>
<tbody>
<tr>
<td>17. Jeg våkner ofte om natten.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>18. Jeg sover godt hele natten.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>19. Jeg er ofte koptlustet.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>22. Jeg er tungpusten når jeg snakker.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>23. Det anstrenger meg veldig å få besok.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>24. Jeg hoster mye.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>25. Jeg har ofte slim i luftveiene.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>27. Jeg føler meg vel sammen med venner og bekjente.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>28. Jeg er redd for å få anfall med tungpust.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>29. Jeg er tungpusten ved fysisk anstrengelse.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>30. Jeg er irritert over imøkløknin årene som min sykdom medfører.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>31. Min ekstropia påforhold i der under-sykdommen min.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>32. Jeg kan gå og handle.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>33. Jeg kan utøve alle fritidsaktiviteter som interesserer meg.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

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Hvordan har du hatt det i løpet av *den siste uken*? Vennligst sett et kryss ved svaret som passer best med HVERT enkelt utsagn.

<table>
<thead>
<tr>
<th>Stemmer</th>
<th>Stemmer</th>
<th>Stemmer</th>
<th>Stemmer</th>
<th>Stemmer</th>
</tr>
</thead>
<tbody>
<tr>
<td>ikke</td>
<td>nesten</td>
<td>til</td>
<td>ganske</td>
<td>helt</td>
</tr>
<tr>
<td>det</td>
<td>ikke</td>
<td>en viss</td>
<td>bra</td>
<td>hele</td>
</tr>
<tr>
<td>hele tatt</td>
<td></td>
<td>grad 0</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>

| 34. Jeg føler meg ofte irriteret. | | | | |
| 35. På grunn av sykdommen min er kontakten med venner/feiret imot krenket. | | | | |
| 36. Jeg gleder meg over livet mitt. | | | | |
| 37. Jeg kan delta på sosiale sammenkomster. | | | | |
| 38. Jeg er ofte trist. | | | | |
| 39. Pustevanskene mine plager meg når jeg er ute blant folk. | | | | |
| 40. Jeg er ofte nervøs. | | | | |
| 41. Jeg kan kle på meg selv. | | | | |
| 42. Jeg er trett på dagtid. | | | | |
| 43. Jeg føler meg isolert. | | | | |
| 44. Jeg klarer meg fint når det gjelder sykdommen min. | | | | |
| 45. Mine pusteplager hemmer meg i daglige dagse aktiviteter. | | | | |
| 46. Sykdommen belaster familielivet mitt. | | | | |
| 47. På grunn av mine pusteplager har jeg brutt kontakten med andre mennesker. | | | | |
| 48. Mine fridtidsmuligheter er imot krenket. | | | | |
| 49. Generelt er jeg fornøyd med livet mitt. | | | | |

**Tusen takk!**

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Appendix 7

Severe Respiratory Insufficiency Questionnaire

SRI

Spørreskjema om patientens allmenntilstand ved alvorlig respirasjonsinsuffisens

Vurdering

For å gjøre verdiene sammenlignbare oppgis svaralternativene i tall fra 1 til 5:

| Stemmer ikke | => 1 |
| Stemmer litt | => 2 |
| Stemmer delvis | => 3 |
| Stemmer relativt bra | => 4 |
| Stemmer helt | => 5 |

Deretter blir sifrene rekodet, slik at de høyere sifrene skal samsvare med sin verdi:

<table>
<thead>
<tr>
<th>Råverdi</th>
<th>Rekodet verdi</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>5</td>
<td>1</td>
</tr>
</tbody>
</table>

Punkter som skal rekodes:
1, 2, 4, 5, 6, 8, 11, 12, 13, 14,
15, 16, 17, 19, 21, 22, 23, 24,
25, 26, 28, 29, 30, 31, 34, 35,
38, 39, 40, 42, 43, 45, 46, 47, 48;

I neste omgang blir skalaene beregnet. Middelverdiene utregnes dersom minst halvparten av punktene er besvart. Ved hjelp av formlene nedenfor blir råverdiene omgjort til skala verdier mellom 0 og 100.

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Respiratory Complaints

\[ SRI - RC = \frac{\text{Middelverd} i[2,5,12,19,22,24,25,29] - 1}{4} \times 100 \]

Physical Functioning

\[ SRI - PF = \frac{\text{Middelverd} i[1,16,32,33,41,45] - 1}{4} \times 100 \]

Attendant Symptoms and Sleep

\[ SRI - AS = \frac{\text{Middelverd} i[6,9,11,14,17,18,42] - 1}{4} \times 100 \]

Social Relationships

\[ SRI - SR = \frac{\text{Middelverd} i[7,10,21,27,43,46] - 1}{4} \times 100 \]

Anxiety

\[ SRI - AX = \frac{\text{Middelverd} i[8,13,26,28,39] - 1}{4} \times 100 \]

Psychological Well-Being

\[ SRI - WB = \frac{\text{Middelverd} i[4,20,30,34,36,38,40,44,49] - 1}{4} \times 100 \]

Social Functioning

\[ SRI - SF = \frac{\text{Middelverd} i[3,15,23,31,35,37,47,48] - 1}{4} \times 100 \]


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Spørreskjema for bruker av hjelpemiddel til pustestøtte

For hvert spørsmål, vennligst kryss av det svaralternativet som passer best for din situasjon. Hvis du er usikker på hva du skal svare, vennligst svar så godt du kan.

1. Sivilstand (sett ett kryss):
   - Gift / samboer □
   - Enslig □
   - Skilt □
   - Enke / enkemann □

2. Høyeste utdanning (sett ett kryss):
   - Grunnskole □
   - Videregående skole □
   - Høyskole □
   - Universitet □

3. Tilknytning til arbeidslivet (sett ett eller flere kryss):
   - Yrkesaktiv □
   - Uføretrygd □
   - Helt □
   - Delvis □
   - Helt □
   - Delvis □

4. Hvor mange timer i gjennomsnitt pr.døgn bruker du din hjemmerespirator eller din BIPAP? (sett ett kryss)
   - 5-8 timer □
   - 8-12 timer □
   - 12-18 timer □
   - 18-24 timer □

5. Hvordan er du tilkoblet din hjemmerespirator eller din BIPAP? (sett ett kryss)
   - Via en maske □
   - Via tracheostomi (åpning på halsen inn til luftroret) □

Dersom du har tracheostomi kan du kryss av på følgende utsagn:

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt</th>
<th>Stemmer nesten ikke</th>
<th>Stemmer til en viss grad</th>
<th>Stemmer ganske bra</th>
<th>Stemmer helt</th>
</tr>
</thead>
<tbody>
<tr>
<td>-2</td>
<td>-1</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>

6. Jeg fikk tilstrekkelig informasjon før etablering av tracheostomien

7. Jeg har fått tilstrekkelig opplæring vedr. min tracheostomi

8. Jeg har mye ubehag av min tracheostomi
   Nesemaske □ Maske som dekker både munn og nese □ Munnstykke □ Munnmaske □

10. Har du problemer med bruk av masken? (sett ett kryss)
    Ja □ Nei □

11. Hvis JA, hvilke problemer har du? (sett ett eller flere kryss)
    Lekkasje □ Trykksår □ Kondens □
    Annet □
    ........................................................................................................................................

12. Hvor fikk du opplæring i bruk av din hjemmerespirator eller din BIPAP? (sett ett eller flere kryss)
    Sykehus poliklinikk □ Sykehus sengepost □ Seksjon for behandlingshjelpemidler □
    Andre steder □
    ........................................................................................................................................

Kan du kryssse av på følgende utsagn:

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt</th>
<th>Stemmer nesten ikke</th>
<th>Stemmer til en viss grad</th>
<th>Stemmer ganske bra</th>
<th>Stemmer helt</th>
</tr>
</thead>
<tbody>
<tr>
<td>12</td>
<td>-1</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>

13. Jeg fikk tilstrekkelig opplæring i bruk av min hjemmerespirator eller min BIPAP
    ........................................................................................................................................

14. Trenger du hjelp til daglig til bruk av din hjemmerespirator eller din BIPAP? (sett ett kryss)
    Ja □ Nei □

15. Hvis JA, hvem er det som hjelper deg? (sett ett eller flere kryss)
    Personlig assistent □ Ektefelle/samboer □ Barn □ Hjemmesykepleien □
   Andre □
    ........................................................................................................................................
Dersom du har behov for personlig assistent eller hjelp fra hjemmesykepleien kan du krysse av på følgende utsagn:

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt −2</th>
<th>Stemmer nesten ikke −1</th>
<th>Stemmer til en viss grad 0</th>
<th>Stemmer ganske bra 1</th>
<th>Stemmer helt 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>16. Mine hjelpere har fått tilstrekkelig opplæring</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17. Jeg får tilstrekkelig hjelp</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

18. Har du hatt hjemmebesøk av helsepersonell fra spesialist helsetjenesten (personell fra sykehuset)? (sett ett kryss)

Ja [ ] Nei [ ]

19. Dersom ja, hvor ofte?

Ca. 1 gang pr. halvår [ ] Ca. 1 gang pr. år [ ] Sjeldnere enn 1 gang pr. år [ ]

Annet [ ]

Vedr. oppfølging fra spesialisthelsetjenesten /sykehuset kan du krysse av på følgende utsagn:

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt −2</th>
<th>Stemmer nesten ikke −1</th>
<th>Stemmer til en viss grad 0</th>
<th>Stemmer ganske bra 1</th>
<th>Stemmer helt 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>20. Jeg opplever å få tilstrekkelig oppfølging</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Hvordan vil du beskrive din livssituasjon:

<table>
<thead>
<tr>
<th>Stemmer ikke i det hele tatt −2</th>
<th>Stemmer nesten ikke −1</th>
<th>Stemmer til en viss grad 0</th>
<th>Stemmer ganske bra 1</th>
<th>Stemmer helt 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>21. Jeg føler meg trygg i min hjemmesituasjon</td>
<td></td>
<td></td>
<td></td>
<td></td>
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</tbody>
</table>

22. Har du hatt hjelp til å fylle ut spørreskjemaet? (sett et kryss)

Ja [ ] Nei [ ]

_Vennligst se etter at du har svart på alle spørsmål._

_Takk for hjelpen!_