Children living with Home Mechanical Ventilation
The everyday life experiences of the children, their siblings, parents and personal care assistants

Åsa Israelsson-Skogsberg
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Abstract


Aim: The overall aim of this thesis was to explore the everyday life experiences of living with Home Mechanical Ventilation (HMV) from the perspective of the children and their siblings, parents and personal care assistants.

Methods: Study I describes the experiences of personal care assistants (PCA) working with a ventilator-assisted person at home, based on qualitative content analysis according to Elo and Kyngäs (2008), of 15 semi-structured interviews. Study II, using qualitative content analysis according to Graneheim and Lundman (2004), focuses on exploring everyday life experiences from the perspective of children and young people on HMV, by means of interviews with nine children and young people receiving HMV. Study III, using a phenomenological hermeneutical method, illuminates the everyday life experiences of siblings of children on HMV, based on ten interviews. Study IV explores HRQoL, family functioning and sleep in parents of children on HMV, based on self-reported questionnaires completed by 85 parents.

Results: PCAs working with a person with HMV experienced a complex work situation entailing a multidimensional responsibility. They badly wanted more education, support, and an organisation of their daily work that functioned properly. Children with HMV had the feeling that they were no longer sick, which included having plans and dreams of a future life chosen by themselves. However, at the same time, there were stories of an extraordinary fragility associated with sensitivity to bacteria, battery charges and power outages. The siblings’ stories mirror a duality: being mature, empathetic, and knowledgeable while simultaneously being worried, having concerns, taking a lot of responsibility, being forced to grow up fast, and having limited time and space with one’s parents. Parents of children with HMV reported low HRQoL and family functioning in comparison with earlier research addressing parents of children with long-term conditions. One in four parents reported moderate or severe insomnia.

Conclusion: Children receiving HMV may feel that they are fit and living an ordinary life, just like their healthy peers. At the same time the results of this thesis indicate that everyday life in the context of HMV is a fragile construct that in some respects resembles walking a tightrope. The fragility of the construct also affects the everyday lives of the families and the PCAs.

Keywords: Home Mechanical Ventilation, children, siblings, parents, family, personal care assistants, health, family functioning, everyday life

Original papers

This thesis is based on the following papers that will be referred to in the text by their Roman numerals.

Paper I

Paper II

Paper III

Paper IV

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## Contents

<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Original papers</td>
<td>3</td>
</tr>
<tr>
<td>Abbreviations</td>
<td>6</td>
</tr>
<tr>
<td>Introduction</td>
<td>7</td>
</tr>
<tr>
<td>Background</td>
<td>7</td>
</tr>
<tr>
<td>Children and home mechanical ventilation</td>
<td>7</td>
</tr>
<tr>
<td>Medical conditions necessitating HMV treatment among children</td>
<td>8</td>
</tr>
<tr>
<td>Mechanical ventilation</td>
<td>9</td>
</tr>
<tr>
<td>Home mechanical ventilation</td>
<td>10</td>
</tr>
<tr>
<td>Invasive and noninvasive home mechanical ventilation</td>
<td>10</td>
</tr>
<tr>
<td>The concept home – involving medical technology and a workplace for professionals</td>
<td>12</td>
</tr>
<tr>
<td>Swedish regulations and personal care assistance</td>
<td>13</td>
</tr>
<tr>
<td>PCA and the HMV context</td>
<td>14</td>
</tr>
<tr>
<td>To be a family</td>
<td>15</td>
</tr>
<tr>
<td>The concept family</td>
<td>15</td>
</tr>
<tr>
<td>To be a child or adolescent with HMV</td>
<td>15</td>
</tr>
<tr>
<td>To have a brother or sister with a long-term condition</td>
<td>17</td>
</tr>
<tr>
<td>To be a parent of a child with HMV</td>
<td>18</td>
</tr>
<tr>
<td>Theoretical framework</td>
<td>19</td>
</tr>
<tr>
<td>Lifeworld perspective</td>
<td>19</td>
</tr>
<tr>
<td>Daily life</td>
<td>20</td>
</tr>
<tr>
<td>Caring</td>
<td>21</td>
</tr>
<tr>
<td>Health</td>
<td>21</td>
</tr>
<tr>
<td>Quality of life and health-related quality of life</td>
<td>22</td>
</tr>
<tr>
<td>Rationale</td>
<td>23</td>
</tr>
<tr>
<td>Aim</td>
<td>24</td>
</tr>
<tr>
<td>Overall aim</td>
<td>24</td>
</tr>
<tr>
<td>Specific aims</td>
<td>24</td>
</tr>
<tr>
<td>Methods</td>
<td>24</td>
</tr>
<tr>
<td>Design</td>
<td>24</td>
</tr>
<tr>
<td>Content analysis</td>
<td>24</td>
</tr>
<tr>
<td>Phenomenological Hermeneutics</td>
<td>25</td>
</tr>
<tr>
<td>Phenomenology</td>
<td>26</td>
</tr>
<tr>
<td>Hermeneutics</td>
<td>26</td>
</tr>
<tr>
<td>Preunderstanding</td>
<td>27</td>
</tr>
<tr>
<td>A descriptive and comparative study design</td>
<td>28</td>
</tr>
<tr>
<td>Study I</td>
<td>28</td>
</tr>
<tr>
<td>Participants</td>
<td>28</td>
</tr>
<tr>
<td>Data collection</td>
<td>28</td>
</tr>
<tr>
<td>Data analysis</td>
<td>29</td>
</tr>
<tr>
<td>Study II</td>
<td>29</td>
</tr>
<tr>
<td>Participants</td>
<td>29</td>
</tr>
<tr>
<td>Data collection</td>
<td>30</td>
</tr>
<tr>
<td>Data analysis</td>
<td>31</td>
</tr>
<tr>
<td>Study III</td>
<td>31</td>
</tr>
<tr>
<td>Participants</td>
<td>31</td>
</tr>
<tr>
<td>Data collection</td>
<td>32</td>
</tr>
<tr>
<td>Data analysis</td>
<td>32</td>
</tr>
<tr>
<td>Study IV</td>
<td>33</td>
</tr>
<tr>
<td>Participants</td>
<td>33</td>
</tr>
<tr>
<td>Data collection</td>
<td>33</td>
</tr>
<tr>
<td>Data analysis</td>
<td>34</td>
</tr>
<tr>
<td>Ethical considerations</td>
<td>35</td>
</tr>
<tr>
<td>Results</td>
<td>37</td>
</tr>
<tr>
<td>Study I</td>
<td>37</td>
</tr>
<tr>
<td>Study II</td>
<td>38</td>
</tr>
<tr>
<td>Study III</td>
<td>39</td>
</tr>
<tr>
<td>Study IV</td>
<td>40</td>
</tr>
<tr>
<td>Discussion</td>
<td>41</td>
</tr>
<tr>
<td>Methodological considerations</td>
<td>41</td>
</tr>
<tr>
<td>Overall design</td>
<td>41</td>
</tr>
<tr>
<td>Trustworthiness in qualitative research (I-III)</td>
<td>41</td>
</tr>
<tr>
<td>Validity and reliability in quantitative research (IV)</td>
<td>44</td>
</tr>
<tr>
<td>Discussion of the findings</td>
<td>46</td>
</tr>
<tr>
<td>Conclusions</td>
<td>53</td>
</tr>
<tr>
<td>Clinical implications</td>
<td>54</td>
</tr>
<tr>
<td>Future research</td>
<td>55</td>
</tr>
<tr>
<td>Acknowledgements</td>
<td>56</td>
</tr>
</tbody>
</table>
Introduction

Children with home mechanical ventilation (HMV) in several respects constitute an unexplored population within health research. They form a heterogeneous group of children with varying medical diagnoses and a need for HMV. Unique and often advanced care is delivered by parents and personal care assistants (PCA) outside a hospital setting (Brookes, 2019; Edwards et al., 2017; Ramsey et al., 2018; Sterni et al., 2016; Swedberg et al., 2015). This implies life situations where family members and family functioning face challenges, described as a family affair (Falkson et al., 2017; Knecht et al., 2015).

This thesis is constructed on narratives from children receiving HMV, their families’ and PCAs which will hopefully challenge perceptions about what, for many people, is considered as being ill, can also be perceived as being healthy; despite having one or more serious medical diagnoses one can still feel healthy. It is my wish that this thesis will contribute new knowledge about what seemingly can be regarded as divergent can at the same time be perceived as obvious and natural. Being in hospital can feel like being at home; healthcare professionals in the hospital can be perceived as family and favorite friends can be found at the hospital’s play therapy. Children and young people can feel independent and live on their own even though they have two PCAs by their side around the clock.

Background

Children and home mechanical ventilation

A well-known definition of children with HMV treatment is “any child who, when medically stable, continues to require a mechanical aid for breathing, after an acknowledged failure to wean, or slowness to wean three months after the institution of ventilation” (Jardine & Wallis, 1998, p. 762). HMV in this thesis is defined as supported ventilation such as breathing support via a tracheotomy only, tracheotomy in combination with HMV or HMV via a facemask. The main goal of HMV treatment is to reduce the effort the child has to make to maintain their breathing, which in turn promotes growth, provides energy for activities and improves the conditions that will allow them to achieve their developmental potential (American Thoracic Society, 2016; Amin & Fitton, 2003). It is important to consider that these children are fragile and often need complex care - but with committed and well-educated families and PCAs they can attend school and live rich and joyful lives (Brookes, 2019; Rusalen et al., 2017; Seear et al., 2016). A child in this thesis is defined as a person <
18 years of age, according to the United Nations Convention on the Rights of the Child (UNCRC) (UNCRC, 1989) and includes teenagers and young people as well.

The population of children requiring home mechanical ventilation (HMV) has grown in number and complexity since the introduction of the treatment nearly 40 years ago (Amirnovin et al., 2018). There is no exact numbers for children receiving HMV internationally (Chatwin et al., 2015; Chau et al., 2017; Garner et al., 2013; Rose et al., 2015; Wallis et al., 2011), but data from western countries suggest that over the last 15 years the number has increased ten-fold (Brookes, 2019). In Sweden the estimated number is about 300 children (Swedevox, 2019), and gradually increasing, in line with the international trend.

Medical conditions necessitating HMV treatment among children
There are a number of medical conditions that can cause respiratory failure and require a child to use HMV. The most common are weakness in the respiratory muscles, failure in the neurologic ventilation control and various kinds of airway and lung pathologies and chest wall diseases (Amaddeo et al., 2016; American Thoracic Society, 2016; Chatwin et al., 2015; Chau et al., 2017; Chiang & Amin, 2017; Preuthipan, 2015). Common diagnoses are neuromuscular diseases, for example Spinal muscular atrophy (SMA) and Duchenne muscular dystrophy (DMD) (Arens & Muzumdar, 2010). Spinal muscular atrophy affects the motor nerve cells in the spinal cord, resulting in an inability to walk, eat, or breathe (Boroughs, 2017). DMD is a hereditary disease occurring almost exclusively in boys, where the muscles slowly break down. The first symptoms usually appear at the age of 3-4 years when the child can suddenly start to fall easily, have difficulties in getting up from the floor and a reduced ability to run. Current improvements in care strategies for children with DMD, including symptom treatment with corticosteroids, treatment of cardiomyopathy and physiotherapy may delay the need to start HMV treatment (Chatwin et al., 2015; Yiu & Kornberg, 2015). Boys with DMD were among the first groups to receive non-invasive HMV and they represent one of HMVs greatest success stories (Simonds, 2016). The HMV treatment has been shown to increase survival; children with various kinds of muscular diseases now have a significant probability of reaching adulthood (Amaddeo et al., 2016; Chatwin et al., 2015; Simonds, 2016).

Central hypoventilation syndrome (CCHS) represents a failure in the neurologic ventilation control. It is a rare genetic disease in which the child stops breathing when falling asleep and ventilator support is a cornerstone of CCHS management (Verkaeren et al., 2015).

Mechanical ventilation
Breathing is one of our most fundamental rhythms connecting us to life; a dimension of the self, our thoughts, feelings and moods which we can only partially control. Respiration is necessary to sustain life; it is an absolute human need, controlled by chemical regulation and the nervous system. The respiratory control system aims to satisfy metabolic requirements. The structure allows breathing to provide oxygen, remove carbon dioxide and regulate hydrogen ion concentration in blood and body tissues. A respiratory system with intact structures and functions is necessary for optimal gas transport (Morton & Fontaine, 2013). If there is an imbalance in the respiratory system, for instance due a lack of respiratory muscle strength, reaches a certain threshold, alveolar hypoventilation occurs (Amaddeo et al., 2016). Respiratory discomfort is a complex, debilitating and multidimensional sensation that may prevent mobility, cause social constraint and shrink a person’s world (Haugdahl et al., 2017).

The modern era of mechanical ventilation developed most noticeably as a response to the polio epidemic that erupted worldwide both through the late 1920s and into 1950s. The iron lung, with negative-pressure ventilation, was developed and used to treat and maintain life for those whose breathing capability had been impaired or destroyed in the wake of poliomyelitis. The whole body was enclosed in the device which formed an airtight chamber. Only the head protruded from the device, tightly sealed around the neck. Positive pressure invasive ventilators became available in the 1940s and 1950s (Dunphy, 2001; Kacmarek, 2011; Lewarski & Gay, 2007). The historical development of mechanical ventilation is remarkable and reflects the evolution of both respiratory and critical care (Kacmarek, 2011).
Home mechanical ventilation

HMV as it exists today can be traced to the early to mid-1980s when tools were developed to manage the care of people suffering from breathing difficulties outside a hospital (King, 2012; Lewarski & Gay, 2007). Advances in pediatric and neonatal intensive care and the development of HMV treatment, with several choices of sophisticated ventilators available, opened the door for both adults and children in need of respiratory support to live in their own homes (Amaddeo et al., 2016; Amirnovin et al., 2018; Brookes, 2019; Castro-Codesal et al., 2017; McDougall et al., 2013; Preuthipan, 2015; Simonds, 2016; Wallis et al., 2011). The development of small and smart HMV devices with durable modern batteries allows children with HMV today to live an active mobile life (Seear et al., 2016). It is extremely important that these devices are properly designed (Fierro & Panitch, 2019; Fu et al., 2012; Lang, 2010) and equipped with adequate audible alarms to alert caregivers in time (Swedevox, 2019).

Invasive and noninvasive home mechanical ventilation

HMV can be delivered invasively via tracheotomy, in some cases continuously as life supportive assistance for 24 hours, or noninvasively through a face mask, typically during sleep (Chatwin et al., 2015; King, 2012; Simonds, 2016). Treatment strategies are determined by the individual pathophysiological respiratory failure (Amaddeo et al., 2016). There is a wide variety of indications for treatment and consequently very different outcomes for each child. In a Swedish study (Laub et al., 2006) the most common reason for starting HMV among children with SMA was cough insufficiency. Some children might receive both NIV and invasive treatment during their lifetime, depending on their underlying conditions. Some children can be weaned from the HMV treatment when their condition improves with growth and development (Brookes, 2019), but HMV is not in itself a curative treatment (Edwards et al., 2017).

Tracheotomy

Tracheotomy is a surgical and operative procedure where an opening is made on the front of the throat to create a free airway (Chiang & Amin, 2017; Morton & Fontaine, 2013). A tracheostomy tube is inserted into the trachea and the ventilator is attached by flexible tubings. This invasive treatment is more commonly used when the child needs the ventilator continuously (American Thoracic Society, 2016), usually when HMV support is required for > 16 hours a day and cannot be managed with NIV. This can be the case, for example, if oral secretions or uncontrollable gastroesophageal reflux disease excludes the safe use of NIV (Amin et al., 2017). According to data from Swedevox (2019) the main diagnoses for children treated with tracheotomy in Sweden are: unspecified diagnosis (25%); neuromuscular disease and polyneuropathies (18%); brain damage (congenital or acquired) (13%); bronchopulmonary dysplasia (BPD) (13%).

This is a safe method for delivering long-term ventilation but airway emergencies do happen, and a small child with a tracheotomy should not be left alone because of the risks associated with ventilator disconnection or mucous plugging (Chiang & Amin, 2017). A tracheotomy has to remain in place, stay open and free from mucus (American Thoracic Society, 2016; Chiang & Amin, 2017; Hanks & Carrico, 2017; King, 2012; Lewarski & Gay, 2007). Both invasive and non-invasive HMV treatments are complex respiratory services to organise in a home setting and require the provision of special care and training. Some children with tracheotomy can manage without ventilator support during the day and a speaking valve can be connected to the tracheotomy (American Thoracic Society, 2016). Consequently, this is a diverse group of children and families with a unique need for support if they are to achieve an everyday life that functions well.

Noninvasive ventilation; CPAP and BiPaP

If ventilator support is required only during sleep, a more noninvasive approach is taken. This treatment involves a mask placed on the nose or face to provide assistance with breathing. It is used increasingly worldwide in children of all ages (Brookes, 2019). There are two types of NIV; continuous positive airway pressure (CPAP) and biphasic positive airway pressure (BiPaP). CPAP delivers a constant positive pressure and aims to maintain an open airway throughout the breathing cycle. In children, CPAP is commonly used for the treatment of sleep apnea syndrome, muscular hypotension, craniofacial malformations, obesity and tonsil surgery with suboptimal results (Brockbank, 2017; Swedevox, 2016). BiPaP delivers a
supplemental higher positive pressure during every inspiration to assist breathing (Amaddeo et al., 2016; Chiang & Amin, 2017; Grychtol et al., 2018).

The concept home – involving medical technology and a workplace for professionals

The advancement of medical technology during the last two decades has allowed children with complex medical needs to leave hospital and remain in their own homes with their families (Brookes, 2019; Chatwin et al., 2015). A home can be understood as a person’s innermost room; a room where we exist, act together and shape our identity (Hilli & Eriksson, 2017). It is a private sphere of integrity and relates to concepts such as fellowship, relationships, caring and security and has been described as a sacred place in a profane world (Liaschenko, 1994). Martinsen (2006) writes about a house that sings; one where the inner is in harmony with the outer. A house that sings is a secure place on earth where we can dwell and feel a sense of belonging. Individuals can also feel homeless in their own home if the atmosphere is negative and the special feeling about being home is lost (Hilli & Eriksson, 2017). The feeling of being safe, centered and connected seems to comprise essential aspects of the feeling of at-homeness (Ohlen et al., 2014; Tryselius et al., 2018). Home in this thesis is interpreted as the private place where there is space for everyday life and personal growth; a place in which to find peace, memories, rest and the possibility of living according to one’s own habits and wishes, with friends and family (Lindahl et al., 2011). A place where it is possible to develop one’s own identity and feel safe (Andersson et al., 2019).

Nonetheless, children with HMV and their families must sometimes spend long periods in hospital. The concept of home might then take on several meanings; it could indicate the “real home” or the family accommodation near the hospital, without either one being the obvious favourite. To feel at-homeness could include being in the hospital’s playground, in a familiar hospital ward or in the family accommodation near the hospital together with the family. One important aim for professional caring may therefore be regarded as creating places and spaces (Lindahl, 2018; Lyckhage et al., 2018; Tryselius et al., 2018) in this context, where children with HMV and their families can dwell and feel secure.

Children receiving HMV may, in addition to breathing difficulties, have severe functional limitations (Amin et al., 2017; Brookes, 2019) resulting in their home frequently becoming a place containing a lot of medical technology. When advanced medical technology is introduced into a private home, into a room that used to be a private space and not designed for the receiving or providing of healthcare, it may engender changes requiring adaptations that might affect the family’s options regarding autonomy (Lang et al., 2014; Lindahl & Kirk, 2018; Lindberg et al., 2016).

In Sweden children receiving HMV treatment are regularly supported by PCAs at school and at home, in partnership with parents during the day and at night. This is an intimate work situation characterised by the PCA’s constant presence in the family’s home (Swedish National Board of Health and Welfare, 2005). This relationship can last for many years (Martinsen et al., 2018) and there might be difficulties in establishing boundaries, in terms of work and leisure, friendship and a professional relationship (Swedberg et al., 2013). Tensions may arise, as this arena is both a home and a workplace (Ahstrom & Wadensten, 2012) with different meanings for the owner and the guest. It is not necessarily suitably furnished as an environment for the provision of care with appropriate working conditions, which calls for a humble attitude on the part of all those involved (Martinsen et al., 2018).

Swedish regulations and personal care assistance

Sweden has ratified the United Nations Convention on the Rights of Persons with Disabilities (CRPD) and Swedish disability policy is founded in human rights. The act “Support and Services to Persons with Certain Functional Impairments” (LSS 1993:387), came into force in Sweden in 1994 (Swedish National Board of Health and Welfare, 2007). This act regulates PCA and aims to promote independence, individual support and participation in society for a group of people with defined disabilities. The ethical perspective that influenced the act is that all people are equal; no human being is worth more than another, and everyone has the right to integrity and privacy (Brusén & Flyckt, 2013; Clevnert & Johansson, 2007; Lewin, 2011; Olin & Dunér, 2016).

In order to be eligible for personal care assistance, one must have fundamental and basic needs for assistance in daily life. The cost of PCA is split between the state and the municipality. When the assistance is required for 20 hours a week or less the municipality cover the cost, while the state is responsible when the need exceeds 20 hours a week (Swedish National Board of Health and Welfare, 2007). Personal care assistance has been the subject of much debate in Sweden over the last few years following sharp increases in the cost.
This has led to political decisions limiting the allowance for PCA support for children on HMV. This uncertain situation has seriously affected the families concerned, and has been reported on by Swedish television, newspapers and social media.

PCA is provided by the municipality, a private assistance company, a co-operative or even family member (Clevnert & Johansson, 2007; Dunér & Olin, 2018; Olin & Dunér, 2016; Swedish National Board of Health and Welfare, 2007). Consequently it is possible for parents to work as a PCA for their own child (Swedish National Board of Health and Welfare, 2007). Many parents in Sweden, in contrast to many other countries, are employed as paid PCAs for their children (Olin & Dunér, 2016). This might be challenging; handling multiple roles may end in a situation where the work role and the parental role become blurred (Ranehov & Hakansson, 2018).

**PCA and the HMV context**

PCAs have different degrees of professional training and varying levels of education (Schaepe & Ewers, 2017; Swedberg et al., 2013). There are no formal competence requirements for working as a PCA in Sweden. Education and delegation of care to PCAs in the HMV context requires stringent organisation to ensure good and safe care. Healthcare professionals often train PCAs in specific care activities, such as cannula care and respiratory management, since each child’s needs are very diverse and unique (Astrid Lindgrens Barnsjukhus, 2016; Swedberg, 2014; Swedevox, 2016).

Research in this area emphasises that PCAs are often left alone with responsibilities that far exceed their level of knowledge and training (Swedberg et al., 2013; Ward et al., 2015). They must have a high degree of confidence in their skill to cope with an airway or ventilator emergency (Amin et al., 2017; Ramsey et al., 2018). Parents, who often become experts in the management of their child’s condition (Whiting, 2013), acquire a high level of knowledge, skill and competence from PCAs working with their child. Tensions may arise if parents find the PCA’s skills, knowledge, level of confidence and self-assurance to be suboptimal and therefore lack confidence or trust in them (Amin et al., 2017; Maddox & Pontin, 2013; Ward et al., 2015). The opposite can also happen; parents tell the PCAs that their treatment is incorrect, despite that the PCAs following agreed care protocols (Maddox & Pontin, 2013). Nevertheless there are also reports (Ahlstrom & Wadensten, 2011; Amin et al., 2017) of parents’ appreciating and feeling blessed with PCAs whose care is grounded in dignity and empowerment. More knowledge is needed about PCAs’ needs and requests for adequate training and support to allow them to feel safe and secure at work.

**To be a family**

**The concept family**

Family as a concept is understood in this thesis to denote a unit where all members have a part, which taken together results in something larger than the sum of the parts. Each family member is both a part and a unit in themselves. The family together forms a functional unit where the interactions can be described as family functioning, which embodies the characteristics of the family (Wright & Leahey, 2013). Six major dimensions of family functioning have been identified; communication, problem-solving, roles, affective responsiveness, affective involvement, and behavior control (Miller et al., 2000). Family functioning is an important area of research not least in the context of long-term pediatric conditions where family functioning is often affected in various ways (Herzer et al., 2010).

Families have a sense of belonging and share a mutually strong commitment to each other’s life, described as a long-lasting relationship were individuals relate to each other (Wright et al., 2002). Families are shaped in diverse ways with a variety of combinations (Benzein et al., 2012); families can include several generations or be extended families where parents are separated but linked by the child and new mothers and fathers are included, described as multiple households (Ganong, 2003). Wright and Leahey’s (2013, p. 60) inclusive and extensive definition “family is who they say they are” allows people who neither share a household nor are related by blood to constitute a family. Each individual decides who is a family member (Benzein et al., 2008) which is a suitable definition in a HMV context where families to some extent include PCAs and other healthcare professionals.

**To be a child or adolescent with HMV**

Living with HMV can be a lifelong process affecting diverse dimensions of everyday life. Some children may be gradually weaned from the HMV treatment and some possibly from invasive treatment to NIV treatment; for some the need for HMV will increase over time (Brookes, 2019). A metasynthesis study (Lindahl & Lindblad, 2011) emphasised that some children with HMV expressed hopes of giving up the ventilator, but overall, treatment and technological devices were viewed as good and easily managed, creating energy and allowing them to do favorite things. Dreyer et al. (2010); Gibson et al. (2012); Kirk (2010) have
reported that HMV extended the user’s lifespan, and gave them the capacity to live an active life.

Nevertheless, needing HMV regularly includes having a progressive disease where the children and teenagers affected gradually become more dependent and may lose their ability to breathe, walk, get dressed or eat by themselves. This loss creates a growing dependence on others in daily life, at a time when most teenagers are expected to grow, and probably want to become more independent (Pehler & Craft-Rosenberg, 2009; Waldboth et al., 2016). Being a teenager and having Duchenne muscular dystrophy (DMD) has been described as a difficult, worrying and lonely period with reflections about social life and sexuality, and an inability to participate in ‘the wild teenage life’ because of the physical impairment (Dreyer et al., 2010). Pehler and Craft-Rosenberg (2009) have also described teenage longings for relationships. Dependence on others and being unable to participate in activities can complicate developing and maintaining friendship. Björquist et al. (2015) interviewed teenagers with cerebral palsy who told about the importance of participating in activities with friends, experiencing love, managing daily activities and having supportive surroundings. Participation in activities was seen as a challenge because of the need for support. Not being able to participate in the same activities as their friends, made computer games and social media important. Buchholz et al. (2018) have described the importance of being in control of one’s own social network for those with communication disabilities. Online gaming allowed the participants in Buchholz et al. (2018) study the possibility to interact with others and be perceived as just anyone else in the group.

The prominent figures for the attachment theory described by John Bowlby (2010) and Mary Ainsworth (Broberg et al., 2006) maintain that early relationships serve as templates which are important for self-perception in later relationships; internal working models are based on and shaped by previous relationship experiences (Berk, 2007; Bowlby, 2010; Broberg et al., 2006; Broberg, 2015). The Russian phycologist Lev Vygotsky, made an important contribution to the sociocultural theory emphasising the necessity of social interaction for children to acquire thinking and behavior that constitute culture in a society. Vygotsky believed that a society selects important tasks for its members who, through social interactions can gather those competences, which are essential for participation in the society. Adults and skilled peers help children to master cultural activities, and this communication becomes a part of the children’s thinking (Berk, 2007).

Today, research suggests that children's development is a result of biological, social and psychological components affecting each other. These in turn are based on major theoretical perspectives deriving from Sigmund Freud, Erik Homburger Eriksson, Jean Piaget and John Bowlby (Broberg, 2015). Various aspects of children’s development become central at different ages. Children between the ages of three and four can be extra sensitive to separations. A five-year-old child may be afraid of various things concerning death, and questions about why one has to die can arise. Peer relations become increasingly important at school age, where a socialisation process without parental involvement often begins. These relationships are important for perceived self-esteem. A nine-year-old child can have advanced thinking processes and perspectives which imply an ability to see themselves and their family through the eyes of others. In adolescence, it is common to be dissatisfied with one’s appearance and possibly to struggle to accept that it does not match one's ideals. Adolescence often includes a period of trying to find a balance between separation and affinity, and reflections on who you are and what you want to do with your life (Berk, 2007; Broberg, 2015). Many of the young men with HMV and DMC in the Dreyer et al. (2010) study felt independent and that they led active extrovert lives despite high levels of physical dependency in everyday living. They wanted to learn, travel and participate in sport, they had dreams about life just like everyone else. Kirk (2010) has also described children with HMV telling about social activities such as sports- and gaming and going to pubs and clubs. They incorporated the technology into their social and personal identities. Identity development is part of growing up for all young people, and this group of children and teenagers had the additional job of incorporating their illness and technology into the process. There are only a small number of studies that include the voices of children with HMV; yet listening to these voices is a prerequisite for gaining the knowledge needed to create quality care for support in their everyday lives.

To have a brother or sister with a long-term condition
Sibling relationships can be the longest and closest relationships we have in life (Knecht et al., 2015; Meltzer & Kramer, 2016), given rather than earned (Cicirelli, 1995) and constituting an important part of each other’s social environment involving both social support and challenges (Von Tetzhner, 2016). They include conflicts contributing to the development of social and emotional skills and behaviour (Von Tetzhner, 2016; Woodgate et al., 2016).

Pediatric disability and long-term illness often have a significant effect on healthy siblings (Caspi, 2011; Hartling et al., 2014; Knecht et al., 2015; Limbers & Skipper, 2014) with both
positive outcomes - such as satisfactory self-esteem, social resilience and maturity (Barr & McLeod, 2010; Gan et al., 2017) and negative outcomes. Regular visits to the sibling in hospital, increased responsibilities at home and the parents’ reduced capacity to provide the siblings with attention may contribute to stress and anxiety (Gan et al., 2017). There may also be worries about the sick siblings’ ability to take care of themselves in the future (Waldbeth et al., 2016). Siblings may modify their behavior to meet the family’s needs and take on tasks and skills that include them in the family’s current caring goal, behaviour which is finally internalised into their role and identity (Deavin et al., 2018). Previous research has observed that routines around the clock for a child with HMV influenced everyday lives of the other sibling’s (Lindahl & Lindblad, 2011). More knowledge is needed from the siblings’ own perspective on experiences of being a brother or sister of a child with HMV.

To be a parent of a child with HMV

Parenting, as described by Bowlby (2010), includes being available, providing a safe and secure base which a child can leave and return to. Bowlby exemplifies the parental role with the metaphor of being a basecamp commander, sending out an expeditionary force that can return if faced with adversity. If there is a safe base to return to, the forces are capable of progress and take risks. If parents are to remain a safe base, time and a permissive atmosphere is required (Bowlby, 2010).

Parents of children with HMV deliver a wide range of unique, advanced and complex care outside the hospital setting (Brookes, 2019; Edwards et al., 2017; Sterni et al., 2016) which often entails maintaining high levels of vigilance and skilled care, day and night (Keilty et al., 2015; Lindahl & Lindblad, 2013; McCann et al., 2015). This situation may not only change the role of the parents within the family but also affects their function (Falkson et al., 2017; Kirk et al., 2005; Meltzer & Booster, 2016; Wang & Barnard, 2008) as they constantly have to learn and manage new roles (Carnevale et al., 2006). Disturbed nocturnal sleep is common, with mothers in particular reporting poor quality of sleep (Cadart et al., 2018; Nozoe et al., 2016). Parents often go to sleep prepared to respond to alarms from medical devices and wake up early to support the various routines (Heaton et al., 2006; Keilty et al., 2015). Disturbed sleep affects multiple domains of the parents’ perceived health-related quality of life (HRQoL) (Meltzer et al., 2015). It is important to take note of this, (Keilty et al., 2018) since there is an inseparable link between parental HRQoL and the wellbeing of children on HMV (Cadart et al., 2018; Graham et al., 2014; Seear et al., 2016).

Parents’ experiences reported in previous research in the context of HMV tell of a daily life involving both distress and enrichment (Carnevale et al., 2006; Ward et al., 2015), and a loss of privacy at home (Falkson et al., 2017). Positive effects for the whole family have also been reported, in the form of engendering a sense of compassion and understanding for diversity (Mah et al., 2008). Parenting may not decline when the child becomes an adult and the need to be responsible for and watch over their child may last their whole life (Jeppsson Grassman et al., 2012; Yamaguchi et al., 2019). This kind of involvement has been described as “it feels like my son’s disability is under my skin” (Whitaker, 2018, p. 218). There is a shortage of research in this area in Sweden. Several aspects, for instance HMV mode, defined as 1) tracheotomy, 2) non-invasive ventilation (NIV) or 3) continuous positive airway pressure (CPAP), and unique care situations, have not been explored in relation to parental sleep and the impact on HRQoL and family functioning. This ought to be investigated, and measures should be taken to reduce parental strain and exposure.

Theoretical framework

Lifeworld perspective

The qualitative focus of this thesis is on children with HMV, their families’ and PCAs’ everyday lives, including how they experience their health, their values and the meaning of life. Life itself, i.e. lived experiences is the starting point. The creation of human experience with its beauty, emotional actions, favourite music and literature, flavours, sounds and colours is what Husserl called the lifeworld; a world where all meaning originates (Bengtsson, 2011; Galvin & Todres, 2013; Todres et al., 2007; van Manen, 2016). van Manen (1997, p.36) refers to Dilthey who wrote “lived experiences are to the soul what breath is to the body, just as our body needs to breathe our soul requires fulfillment of its existence, like an echo of our inner emotional life”.

It is challenging to describe a world that is lived. van Manen (2016) describes five fundamental lifeworld themes, or existentials. They are universal themes of life that may guide reflections on human experiences regarding HMV treatment; lived time (temporality), lived space (spatiality), lived relation (relationality), lived body (corporeality) and lived technology (materiality).

In the HMV context time that is lived (temporality) (van Manen, 2016) can constitute feelings of possibilities for the future or a future with only limited possibilities (Galvin and Todres,
Lived space (spatiality) is space that is felt, which in this context could be a matter of living in worlds; an inner and an outer world (van Manen, 2016). Relations that are lived are maintained with others, in a social world, via a capacity for language. It includes how we are a part of a tradition or a culture. If we want to understand illness, as it is lived, we have to understand what it means culturally and interpersonally (Galvin & Todres, 2013).

A body that is lived (corporeality) refers to the phenomenological fact that we are bodily involved in the world. Merleau-Ponty (2003/1945) developed the concept and invented the term embodied. He meant that instead of being an object for the world the body becomes the point of departure against the world. Galvin and Todres (2013) argue that illness is not only a physiological explanatory models - it includes the body functioning in a meaningful way in the world. Gadamer (1996) described this as the lived body making itself heard and being aware of being ill. Illness often changes the perception of the body and the relationship with the world.

It is useful to understand the technology as lived when describing the HMV context. A critical and objectifying gaze can mediate feelings of embarrassment and even shame; when our body is seen (and maybe experienced) as an object separated from ourselves feelings of objectification and alienation can emerge (Finlay, 2011). Kirk (2010) describes how children equipped with various forms of medical technology became accustomed to the presence of technology in their body over time and the device becomes like a physical extension of their bodies. Some children felt that visible (for other people) medical technology affected how they were identified and categorised. The medical technology symbolised their difference from their peers.

Daily life
Schutz (1976) writes about a social world and shared reality that is given to us, into which we are born and grow up. Daily life is not a private world; it is a common world where we share a common sector of space and time. We acquire knowledge about this world that hinders us, or lets us realise our plans and dreams, we have to accept it or modify it – decide if it entails happiness or discomfort. This intersubjective world carries meanings for us all; “experienced by the Self as being inhabited by other Selves, as being a world for others and of others” (Schutz, 1976, p. 20).

Caring
One important point of departure for the work in this thesis was Kierkegaards (1877;1859) way of describing what helping another human being might be; “To help someone, I have to understand more than he does but first and foremost mainly understand what he understands. If I cannot do that, it will not help if I can do more and know more. All genuine helpfulness begins with humility for the one I want to help and thus, I must understand that this is because helping does not want to rule without wanting to serve. If I cannot do this, I cannot help either” (Kierkegaard, 1877;1859). Accepting such a perspective, caring means searching for persons where they are; being a guide and showing a possible direction. Human life can be seen as an infinite wandering; an unlimited search. To perceive caring means to show a possible way. This gives caring an ontological (and true) meaning – where we try to understand the other person’s wishes and desires - and to be helpful to that person in their search for the object of their longing (Eriksson, 2018).

Eriksson (1986) describes caregiving as a natural human behavior; everyone is a natural carer; it is a character trait that makes us human. In a modern society characterised by the use of advanced technology, natural care has often been replaced by professional care. Eriksson describes the concept of self-care as an intermediate care between natural and professional care, where self-care is a kind of support for natural care. Care as entirety can be seen as a balancing act between natural care, self-care and being cared by others (professional care). This balancing act requires a high degree of reciprocity between those involved (Eriksson, 1986). The Norwegian nurse and philosopher Kari Martinsen (Alvsvåg, 2014) gave substance and content to the concept of caring when she formulated the definition that caring involves how we relate to each other and are concerned for each other in daily life. She believes that caring is the most natural and fundamental aspect of human existence. Martinsen (2006) describes professionalism in caring as an ability to be human, allowing the other to emerge, protecting integrity and caring for life with both humanity and technology. She sees silence as a precondition for understanding. Listening is not just hearing, it is allowing what one has heard to be settled. Galvin and Todres (2009) describe an important dimension of nursing as an openheartedness that can be derived from one of Levinas’s central insights; subjectivity arises from an exposure to alterity (the other).

Health
The World Health Organization (WHO) defines health as a multidimensional construct, which includes physical, mental (emotional and cognitive) and social dimensions (World Health
basically experiences meaning and context; a process reliant on human relationships and health-related quality of life (HRQoL) as more of a subdomain (Davis et al., 2006) which has transverses various domains (Davis et al., 2006). QoL is considered a universal construct and normally a possible (Whitaker, 2018) (Lindström et al., 2014).

The starting point of the thesis is to perceive health as a concept that can exist even against a background of several complex diseases (Jeppsson Grassman & Olin Lauritzen, 2018; Lindsey, 1996). The boundaries between health and illness can be provisional, fluid and unstable conditions, characterised by unpredictable processes. Health can exist in different types of gaps; it can be a balancing act between health and illness where one tries to live as normally as possible (Whitaker, 2018). This is in line with Merleau-Pontys’ (2003/1945) theory of a holistic lifeworld with interrelated, interconnected horizons of meanings; near is in relation to far, self is in a relation to other. This relation describes an intimately holistic interconnected human aspect of life, where figure and background are reversible and often shift. In Madjars (1997) words we perceive our body in an unconscious way when we experience health. Health can therefore, be considered as an unconscious body or as biological silence. Gadamer (1996) describes health and illness as a disturbance of one’s freedom - a state that involves a kind of exclusion from life. He characterised health as a riddle or mystery with a hidden character; a state where one feels involved with friends and engaged in one’s everyday life. In Parses (1990, p. 140) words; “Health is how I live my life — my own personal commitment to being the who that I am becoming. Listen to me nurse, when I tell you how I am, and what I will do — since that is how I am going to be me”

Quality of life and health-related quality of life
There is no universally established definition of the term quality of life (QoL) (Bratt & Moons, 2015). It has different meanings for different people (Fayers & Machin, 2016; Karimi & Brazier, 2016), but it is generally conceptualised as a broad assessment of well-being that transverses various domains (Davis et al., 2006). QoL is considered a universal construct and health-related quality of life (HRQoL) as more of a subdomain (Davis et al., 2006) which has also been difficult to define; several definitions can be identified in the literature (Karimi & Brazier, 2016). The concept HRQoL relates to the individual’s multidimensional experience of health and how health is affected by illness and treatment (Davis et al., 2006). Measuring HRQoL aims to systematically describe the individual's experience of health and / or illness. The description ought to be both subjective and dynamic, providing an opportunity for an increased understanding. Achieving this requires the use of an instrument comprising various questions that cover different aspects of the individual's experience in various dimensions (Fayers & Machin, 2016). In this thesis Peds QL Family Impact Module (Varni et al., 2004) has been used to assess the impact a child’s long-term health condition has on the parents. The instrument is constructed in accordance with the WHO definition of health (Varni et al., 2004; World Health Organization, 1948).

Rationale
More children survive severe illnesses today which create new groups of children entering into adolescence and adulthood. The number of children receiving long-term HMV has increased, and the development of smart medical technology allows children with complex medical needs to leave hospital and stay in their own homes together with their families. There is also a lack of care places, not least within the intensive care units (Swedish Intensive Care Registry, 2019). Sweden has the lowest proportion of care places among the developed countries in the Organisation for Economic Co-operation and Development (OECD) (OECD, 2017; Swedish National Board of Health and Welfare, 2018). The home of a child receiving HMV generally becomes a place where advanced and specialised care is carried out, which often influences all family members; the everyday lives of the child with HMV and their siblings, the role of the parents and the work situation of the PCAs. Personal care assistance has been the subject of extensive political debate in Sweden in the last few years arising from a sharp increase in cost which has led to decisions that have affected children with HMV and their families.

There is a lack of research from an insider perspective in the context of HMV in Sweden and this thesis contributes new knowledge stemming from family members’ own voices. These new insights can facilitate our understanding of both the unique knowledge that the families have and of how adjustments to life are influenced by the HMV treatment and furthermore, which adjustments are needed on the part of healthcare professionals. With increased knowledge it is possible to develop caring strategies that include dedicated multidisciplinary
pediatric home ventilation teams who support children with HMV, their families and PCAs. To make this possible we need to identify and understand challenges - but also embrace and protect the strength and unique knowledge that these families have.

**Aim**

**Overall aim**
The overall aim was to explore the everyday life experiences of living with HMV from the perspective of the children and their siblings, parents and personal care assistants.

**Specific aims**
The aim of Study I was to describe PCAs’ experiences of working with a ventilator-assisted person (adult or child) at home.

The aim of Study II was to explore everyday life experiences of children and young people living with HMV.

The aim of Study III was to illuminate the everyday life experiences of siblings of HMV-assisted children.

The aim of Study IV was to explore HRQoL, family functioning and sleep in parents of children receiving HMV in Sweden. A secondary aim was to explore the influence on HRQoL, family functioning and sleep on selected potential determinants.

**Methods**

**Design**
This thesis is based in caring and the human sciences; a research field founded on a humanistic approach which aims to gain knowledge about parts of human existence about which silence reigns, such as wellness, alienation, loneliness, happiness and their characteristics (Dahlberg et al., 2008). The design aims to provide a comprehensive picture of human behaviour and experience. Qualitative and quantitative research methodologies such as interviews and questionnaires have been used to reflect the aims and answer the research questions (Crotty, 2015; Katz et al., 2016; Oiler Boyd, 2001) and to obtain knowledge in this area, both broad and deep, portrayed from various perspectives (Table 1).

<table>
<thead>
<tr>
<th>Study</th>
<th>Design</th>
<th>Participants</th>
<th>Data collection</th>
<th>Analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Qualitative descriptive</td>
<td>15 PCAs working with a person with HMV (adult or child) at home</td>
<td>Semi-structured interviews</td>
<td>Qualitative content analysis</td>
</tr>
<tr>
<td>II</td>
<td>Qualitative descriptive</td>
<td>9 children and young people (aged 7-20) with HMV</td>
<td>Open-ended interviews supported by Photovoice methodology</td>
<td>Qualitative content analysis</td>
</tr>
<tr>
<td>III</td>
<td>Qualitative descriptive and interpretive</td>
<td>10 siblings (aged 7-17) of a child with HMV</td>
<td>Narrative interviews</td>
<td>Phenomenological hermeneutical method</td>
</tr>
<tr>
<td>IV</td>
<td>Quantitative Explorative</td>
<td>Parents (n=85) of children with HMV (n=55)</td>
<td>Self-reported questionnaires</td>
<td>Paired t-test ANOVA X²-test Multiple linear regression analysis</td>
</tr>
</tbody>
</table>

**Content analysis**

Interview data in Study I and II were subjected to qualitative content analysis according to Elo and Kyngäs (2008) (Study I) and Graneheim and Lundman (2004) (Study II). Content analysis is empirically secured, scientific tool which aims to achieve a condensed and broad description of any phenomenon under scientific scrutiny. The method can be used to analyse both quantitative and qualitative data. In the analysis process, words are condensed into fewer content-related categories, and the result is often presented as themes or categories that aim to describe the phenomenon (Elo & Kyngäs, 2008; Hsieh & Shannon, 2005; Krippendorff, 2013). Qualitative content analysis originates from a quantitative approach used in media research (Elo & Kyngäs, 2008; Graneheim & Lundman, 2004; Hsieh & Shannon, 2005; Polit & Beck, 2012) and is not linked to a theoretical starting point, i.e. an epistemological or ontological base (Polit & Beck, 2012; Sandelowski, 2000). Although qualitative content analysis studies differ from other qualitative approaches they may be similar in expression and texture (Graneheim et al., 2017; Sandelowski, 2000).

**Phenomenological Hermeneutics**

Study III used a phenomenological hermeneutical method for interpreting interview texts, described by Lindseth and Norberg (2004) and inspired by the theory of interpretation presented by Ricoeur (1976). Using a lifeworld perspective, the method aims to describe, gain insight into and mediate a deep understanding of what another person’s life situation might be (Lindseth & Norberg, 2004). In Ricoeur’s formulation, interpretation is the link between language and lived experience (Geanellos, 2000), where new modes of being, and new
possibilities of orientating oneself in the world can be revealed. Ricoeur defined hermeneutics as a theory of rules that govern text interpretations; a text possess “surplus of meanings”, allowing the meaning behind the text to be interpreted (Ricoeur, 1976, 1981). The text is autonomous and stands alone; and it is open to more than one interpretation. A text has several meanings, and analysis is a process of moving between manifest and latent meanings where understanding and explanation interact and overlap each other (Lindseth & Norberg, 2004). When the text is created and written down, it is separated from the author, the meaning of the text then appears in dialogue with the reader. The text speaks of possible worlds and ways to orientating oneself in the world (Ricoeur, 1981).

Phenomenology
Phenomenology aims to understand everyday life experiences, where the essence of existence is described and unreflected lived experiences are in focus (Finlay, 2011; van Manen, 1997, 2016). Edmund Husserl, seen as the founder of phenomenology, believed that consciousness always has a direction in a prereflected way. Everything is always perceived as something which means something. He named this our lifeworld [die lebenswelt]; an experienced world of meanings. Husserl emphasised the importance of “going to the things themselves” which has become an important expression of the intention of phenomenology; to return to the world, unreflected, as it is perceived (Husserl, 1995). Thus, when phenomenology is used in scientific studies it is important to question the essence of the phenomenon and what it means. Lived experience is the phenomenological starting and endpoint (van Manen, 1997). This core perspective has to be translated into a suitable research methodology which requires an openness and adherence to seeing things as they are and to understanding lived experiences (Galvin & Todres, 2013; Polit & Beck, 2012). The findings are therefore expressed in a narrative form that will help us to understand unique experiences (Galvin & Todres, 2013).

Hermeneutics
Hermeneutics is a research method based the interpretation of text which aims to convey understanding and meaning (Crotty, 2015; Ricoeur, 1981). A consistent theme is the hermeneutic circle, where understanding is described as an expansion of what is already understood. Through shifting perspective between the parts and the whole a new understanding can be developed (Crotty, 2015). Language is a central aspect of hermeneutics; it is a prerequisite for a historical consciousness, in a process where traditions meet and horizons are united (Ödman, 2017). From a hermeneutic perspective, we understand the world from different starting points, which means that we cannot stand outside ourselves in our understanding and interpretation of the world; interpretations and understanding interact (Ödman, 2017).

Preunderstanding
Within a phenomenological method where we want to describe the world as it is experienced by people, and the question of meaning is paramount (Dahlberg et al., 2008), it is essential to strive to confront the data in as pure form as possible, with an openness which includes to hold back personal beliefs and opinions about the phenomenon. This is an approach that Husserl referred to as bracketing (Crotty, 2015; Polit & Beck, 2012) and for which Dahlberg et al. (2008) introduced the term bridling. Compared to Husserl's concepts that focus on keeping pre-understanding under control, Dahlberg et al. (2008) focus more on creating an open mind that will draw attention to the phenomenon when it shows itself. A phenomenological interview focuses on making the interviewee reflect on the phenomenon. Data are collected through open-ended questions, in a reflective manner.

Ricoeur, unlike Husserl, argues that pre-understanding opens up new horizons for trying to understand and explain, and he considers it impossible to confine pre-understanding within parentheses (Ricoeur, 1981). He believes that we belong to a cultural tradition, to a class, to a history that we can never really reflect upon (Ricoeur, 1981). Gadamer (1997) also regard preunderstanding as a tradition and history that we cannot escape from; something necessary for understanding anything. In a hermeneutic approach to research preunderstanding is consecutively regarded as tradition, our history and as constituting a part of our lifeworld, which creates a history of effect [wirkungsgeschichte]. Being aware of our own history of effect can hopefully lead to achieving an awareness of why some circumstances are noticed and others are overlooked. Only then can things be understood in a new way. Meanings are consequences of the past, tied to the present and carried into the future – a fusion of horizons - meaning that interpretation must be sensitive to the historical context (Crotty, 2015; Dahlberg et al., 2008; Gadamer, 1997; van Manen, 2016). According to Ricoeur (1981) the hermeneutical circle moves from our mode of being – from a subjective level to an ontological plane – beyond the knowledge which we may have, to a mode opened up and revealed by the text.

My own preunderstanding derives basically from my clinical work as an intensive care nurse where I met and communicated with people of different ages and a variety of life situations.
This has given me confidence when meeting the children, families and PCAs in this study. I have not worked clinically in the area of HMV, something that I have frequently reflected on. One positive aspect of this lack of experience is, hopefully, that I have been able to preserve an open mind and relate to the phenomena as they have appeared. Throughout the five years I worked on this thesis I wrote notes after all interviews, meetings, telephone conversations with parents, conferences etcetera. Thoughts and reflections, feelings and ideas often arising from deeply emotional conversations and meetings were recorded in my notebook. My aim was to be reflective (Dahlberg et al., 2008). This type of documentation has also been described as an audit trail (Polit & Beck, 2012).

A descriptive and comparative study design
For Study IV a cross-sectional study was designed, using questionnaires to explore parents’ HRQoL, family functioning and sleep in relation to being a mother or father. The child’s HMV mode, defined as 1) tracheotomy, 2) non-invasive ventilation (NIV) or 3) continuous positive airway pressure (CPAP), and whether the parents were employed as a PCA or in paid work outside home were related to the Family Impact Module and Insomnia Severity Index (ISI) reports.

Study I
Participants
The participants in Study I comprised a purposive sample of 15 PCAs - thirteen women and two men, with working experience in this context of one to seventeen years. They were employed by the municipality, a private company or the person with HMV.

Data collection
Data was collected from November 2010 to March 2011. The 15 interviews formed part of two master’s theses; seven interviews were conducted by the author of this thesis and eight interviews by two specialists in nursing in primary health-care enrolled on a master’s programme.

Information about the study was initially given to the PCAs employers who in turn informed the PCAs about the study. Only after a PCA agreed to participate, were they contacted by the interviewer, and additional study information was provided. The participants decided where to meet; 11 interviews were performed in the participants’ own homes, two in an undisturbed room at the University of Borås and two at the participants' workplace. Except on one occasion when one interviewer met two participants all interviews were conducted with individual participants by one interviewer.

Kvale and Brinkman (2014) describe a thematic and dynamic dimension in an interview study; a theoretical clarification which includes the questions why, what and how. Why include defining the aim of the study, what include acquiring knowledge of the subject to be investigated and how is the dynamic dimension; the interpersonal relationship during the interview. The thematic focus determines which aspects of a topic the questions focus on and which remains in the background. A common thematic interview guide was therefore used with themes relating to responsibility, home and education, to guide the interviews in the same directions to fit in with the study aim. Actual themes were decided after a literature review; i.e. the what of the study were identified. An important aspect regarding the dynamic dimension was to be open and attentive to the phenomena, and to be attentive to our own preunderstanding (Dahlberg et al., 2008; van Manen, 1997). Each interview was recorded and transcribed verbatim by the author who conducted it. The author of this thesis subsequently reassessed all 15 transcribed interviews.

Data analysis
Data was analysed as described by Elo and Kyngäs (2008). The text was transcribed verbatim, read several times in order to familiarise the author with the content, and then divided into sections with similar content. A short word (code) describing the content was written in the right-hand margin. Codes referring to content that was similar were grouped together as subcategories. Subcategories containing similar events were identified by interpreting the data and grouped into higher order categories, to describe and abstract the phenomenon of interest.

Study II
Participants
Health-care professionals in outpatient respiratory clinics at three hospitals in Sweden invited children and families to participate through age-appropriate written and oral information. The author only contacted families after they had agreed to join the study. In total 14 families were invited to participate, five families withdrew from the study and a purposive sample of nine
participants; five boys and four girls, aged between 7 and 20 years, with a median age of 11 years, agreed to participate.

Data collection
Data collection was conducted between November 2015 and September 2016. All participants decided on the setting for the interview and selected a comfortable place to meet. Altogether nine interviews were conducted in a participant’s own home with resulting flexibility regarding conditions, requirements and way of communicating. The narrative interviews were recorded. As the participants’ ages ranged from seven to twenty years, each interview situation was adapted to development level and personal wishes. The interviews all differed from each other; some participants wanted to be free to move around in the house, some wanted to lie down on a sofa and some wanted to sit on the floor in their own room. Being invited like this into a family, the child’s home meant entering right into an everyday life which went on parallel with our conversation. Siblings became upset and cried, dogs barked, horses escaped and visitors arrived. There was a lot of activity going on in the home. Based on the participants’ wishes parents were present in six interviews and a PCA was present on one occasion. Their role clarified, strengthened and interpreted the child's voice which could be very weak and hard to hear.

From the starting point that valuable knowledge about childhood is based on children's own experiences (O’Kane, 2008; UNCRC, 1989) the author explained that the child had unique knowledge about what it means to be a child on HMV treatment. It was emphasised from the start that they were experts on their situation with a unique knowledge and there were no right or wrong answers. The interviews started with the question; “Would you like to tell me who you are and what you like to do?”

Photovoice methodology (Wang & Burris, 1997) worked as an inspiration, boosting the dynamic dimension in the interviews. Prior to our meeting the participants were invited to photograph various things that they regarded as essential in their everyday life using the camera in their mobile phone. These photos were then used to stimulate and facilitate the interview and gave participants the opportunity to initiate the interview by talking about their own photos. In some cases we strolled around the house taking photos with the participants telling stories meantime. After each interview, memos of first impressions were written and contextual data regarding the interview situation were noted.

Data analysis
The data analysis in Study II had to be carried out very carefully as sometimes the children’s voices were frail. The first phase included immersion in the texts, photographs and memos. The interview text was transcribed verbatim which was a slow and meticulous process as in some cases it was very difficult to hear the children's voices because they were affected by a weak musculature. In some cases, children and parents spoke simultaneously and finished each other’s sentences, especially when the child’s own voice was not strong enough to tell their story on their own. The different voices were marked in the transcribed material with great accuracy to ensure that the children’s own voices comprised the main data. Data were analysed via qualitative content analysis according to Graneheim and Lundman (2004). The transcribed texts were divided into meaning units, condensed, labelled, coded, and brought together into subcategories and categories. This process resulted in a collection of similar data, sorted into the same place that is important for determining what is in the data. Finally, overall themes were created. Themes are usually quite abstract; they are always an interpretation, described as a basic topic which the overall narrative is about. A theme is a response to the question how (Graneheim et al., 2017; Graneheim & Lundman, 2004; Morse, 2008).

Study III
Participants
Health-care professionals from outpatient respiratory clinics (as in Study II) invited siblings of children with HMV to participate in the study. The approach was identical to that in study II. Two participants were siblings of the participants in study II, the remaining eight participants had no connection to the participants in Study II. On one occasion two siblings from the same family were interviewed together, due to difficulties in organising several meetings with one family. A purposive sample of 10 siblings: four boys and six girls, three older, six younger, and one twin sibling, with a median age of nine years were included to ensure maximum variation in terms of age and gender. All participants had siblings on HMV with diverse long-term medical diseases, extensive functional disabilities and a comprehensive need for support in their daily lives. The participants were used to PCAs working in their home. It may have been preferable to visit the siblings on different occasions, but this was not realistic since these families have very busy schedules and are spread over a wide geographical area.
Data collection
Data were collected through audio-recorded narrative interviews (Kvale & Brinkman, 2014; Mishler, 1986), flexibly designed according to each participant’s individual preferences about where and when to meet. Usually, the interview took place in the family’s home, a secure and familiar place, but in some cases, it took place in a café or an outpatient clinic.

The siblings were also asked to take photographs with their mobile phone cameras prior to our meeting. These were not interpreted or analysed separately, but were simply intended to facilitate our conversation. This seemed to be a suitable way in which to enhance the child’s level of comfort and establish a relationship between the child and researcher, i.e. the aim was to boost the dynamic component in the interview. As in Study II, the siblings decided whether or not to have a parent present. The older participants chose not to, whereas the younger ones wanted to have a parent with them, or nearby. After the introductions and during the interviews the younger children often chose to draw a picture, present a favorite toy or talk about their photos. The open introductory question was the same as in Study II: “Please tell me who you are and what you like to do”. The narratives of the younger siblings were very concrete and comprised distinct memories of toys, places, and situations they had experienced. The older participants’ narratives included more abstract phenomena such as emotions and empathy.

Data analysis
Data were analysed using a phenomenological hermeneutical method (Lindseth & Norberg, 2004) inspired by the French philosopher Ricoeur. The process of analysis involves going back and forth between a naïve reading, one or several structural analysis and a comprehensive understanding with a dialectic movement between understanding and explanation. A naïve reading aims to grasp a first understanding of the meaning as a whole which serves as a starting point for the second steps of interpretation: a distancing and explanatory thematic structural analysis. Ricoeur (1976) explains the structural analysis as a stage between a naïve interpretation and a critical interpretation which makes it possible to locate explanation and understanding. This is a methodical instance of interpretation aimed at identifying and formulating themes. Narratives describing similar meanings are clustered and subthemes and themes are created. In the third and final step of the analysis a comprehensive understanding is formulated, the interpreted whole of the phenomenon, where all the parts of the analysis process are considered and presented in a new whole (Lindseth & Norberg, 2004).

Study IV
Participants
Children with HMV in Sweden are usually treated in respiratory clinics, which in this study were identified via the Swedish National Quality Register for Oxygen and Home Respiratory Treatment (Swedevox). The relevant clinics were informed about the study and healthcare professionals invited parents consecutively to participate between December 2016 and December 2018. In total 88 parents of 55 children; 0-18 years of age receiving HMV, were included. The questionnaires were completed by 85 participants. Paired data, from both mothers and fathers, were obtained from 30 families.

Data collection
Data were collected via Demographic data, Peds QL Family Impact Module (Varni et al., 2004) and Insomnia Severity Index (ISI) (Bastien et al., 2001). The questionnaires were individually completed by each parent, during a hospital visit and placed in a sealed box.

Peds QL Family Impact Module
Disease does not exist in a vacuum; long-term childhood illness and disease influence the sibling bond, the parent-child relationship and the family as a whole (Knecht et al., 2015; Wittenberg & Prosser, 2016). Peds QL Family Impact Module was developed to assess the impact a child’s chronic health condition has on the parents (Varni et al., 2004) regarding HRQoL and family functioning. In the questionnaire HRQoL is defined as a multidimensional construction, which includes physical, mental (emotional and cognitive) and social dimensions in accordance with the World Health Organization (WHO) definition of health (Varni & Limbers, 2009; World Health Organization, 1948). These multiple domains allow the instrument to capture various ways in which parents of children on HMV are impacted (Medrano et al., 2013; Varni et al., 2004). Family Functioning is measured through the items daily activities and family relationships.

The questionnaire consists of 36 items. Three summary scores can be derived: the Total Score (36 items), the Parent HRQoL Summary Score (20 items) and the Family Functioning Summary Score (8 items). The Parent HRQoL Summary Score is calculated by averaging the items from four scales measuring: Physical functioning (6 items), Emotional functioning (5 items), Social (4 items) and Cognitive functioning (5 items). The Family Functioning Summary Score is a summative average of the scales: Daily activities (3 items) and Family relationships (5 items). The Total Score is calculated by averaging the above 28 items with items from the Communication scale (3 items) and Worry scale (5 items). The response
format is a five-point Likert scale where items are scored from “never a problem” (0) to “always a problem” (4) and then reversed and transformed to a 0-100 scale where higher scores indicate better functioning.

The validity and reliability are well-documented (Jastrowski Mano et al., 2011; Medrano et al., 2013; Panepinto et al., 2008; Scarfelli et al., 2008; Varni et al., 2004). The Family Impact Module is translated into Swedish; a linguistic validation was carried out with 109 participants. Subscales were assessed for internal consistency reliability using Cronbach’s alpha. The Total Score, Parent HRQoL Summary Score and Family Functioning Summary Score achieved values greater than 0.70 (Tiberg & Hallstrom, 2009). The questionnaire has been used earlier in Swedish research (Gårdling et al., 2018; Jonsson et al., 2015; Jönsson et al., 2016).

Insomnia Severity Index
The Insomnia Severity Index (ISI) (Bastien et al., 2001) is a seven-item self-reported questionnaire with documented reliability and validity for detecting insomnia (Bastien et al., 2001; Morin et al., 2009; Morin et al., 2011). ISI evaluates sleep-onset (initial), sleep maintenance (middle) and early morning awakening problems (terminal), as well as sleep satisfaction, interference with daily functioning, noticability of sleep problems and distress caused by sleep problems – over the last two weeks. The response format is a five-point Likert scale from “never a problem” (0) to “always a problem” (4). The scores are merged into a total score of 0-28 where a score ≥15 indicates clinically significant insomnia (Bastien et al., 2001). ISI has been used in population studies nationally (Jansson-Frojmark et al., 2016) and internationally (LeBlanc et al., 2007; Morin et al., 2009).

Data analysis
Descriptive statistics was used to present parents’ and children’s background characteristics. Parametric methods were used for analyses after the data were tested for normality and criteria were met. The Paired t-test for comparison of mean values from Peds QL, Total score, Parent HRQoL Summary Score, Family Functioning Summary Score and ISI score were used to analyse differences between mothers and fathers living as couples (n=30). ISI reports were dichotomized, <15 and ≥15, according to the cut-off values defined by Bastien et al. (2001). The associations between the reports of mothers and fathers from ISI were analysed using the chi-square test in relation to HMV mode, working as PCA or not (Graham et al., 2016; Meltzer et al., 2010; Meltzer et al., 2015), parent having paid work outside the home or not (Graham et al., 2016; Navarro Tapia et al., 2018; Secar et al., 2016), HMV mode (Cadart et al., 2018; Graham et al., 2016) and ISI total score (Meltzer and Booster, 2016; Paddeu et al., 2015). A minimum of 90 cases was considered sufficient for the five independent variables (Tabachnick & Fidell, 2014). The adjusted R² evaluated the regression models. A significance level was set to p <.05. IBM SPSS™ Statistics version 25 was used for the analyses.

Ethical considerations
Sweden ratified UNCRC 1990, which emphasise children’s right to dignity, respect, and participation and the importance of letting children’s voices to be heard (Article 12) (UNCRC, 1989). There is a proposal to incorporate UNCRC into Swedish law in 2020 (Government Offices of Sweden, 2016a, 2017) which means there will be a legal responsibility to listen to the voices of children. This thesis steams from children and young people being actors in their own right rather than being seen as pre-adults becoming (Scott, 2008). Their childhood is assigned its own value and is not regarded merely as a preparation for adulthood but rather as part of life which has a value in itself (O’Kane, 2008; Qvortrup, 1994).

This thesis received prior approval from the Regional Ethics Review Board of Gothenburg, Sweden (Ref. No. 354-15), and has been carried out in line with the World Medical Association’s Declaration of Helsinki (WMA, 2013). All research has ethical implications and these studies ethical challenges have continuously been reflected on. Children are vulnerable and less able than adults to assess the consequences of the research they are asked to participate in (Hewitt-Taylor and Heaslip, 2012). Therefore, research that includes children carries a particular ethical responsibility, and risks and benefits must be assessed constantly (Alderson, 2007). The four ethical principles; autonomy, beneficence, non-maleficence and multiple regressions with backward selection were used, after the criteria (normality, linearity and homoscedasticity) were controlled for and satisfactorily met (Tabachnick and Fidell, 2014). The relationships between the dependent variables, HRQoL Summary Score and Family Functioning Summary Score, with the independent variables were predicted. A literature review along with clinical experience prepared the choice of the independent variables; gender (mother or father) (Nozoe et al., 2016; Toly et al., 2012), parent working as a PCA or not (Graham et al., 2016; Meltzer et al., 2010; Meltzer et al., 2015), parent having paid work outside the home or not (Graham et al., 2016; Navarro Tapia et al., 2018; Secar et al., 2016), HMV mode (Cadart et al., 2018; Graham et al., 2016) and ISI total score (Meltzer and Booster, 2016; Paddeu et al., 2015). A minimum of 90 cases was considered sufficient for the five independent variables (Tabachnick & Fidell, 2014). The adjusted R² evaluated the regression models. A significance level was set to p <.05. IBM SPSS™ Statistics version 25 was used for the analyses.
justice have guided all four studies (Beauchamp & Childress, 2013). Study I did not need the approval of an Ethics Committee as health-care professionals are automatically considered to consent to participating in research (Swedish Research Council, 2017).

In order to meet the principle of autonomy age-appropriate written and verbal study information was given to the participants, children and adults alike. Participation in the study was voluntary and could be withdrawn at any time, confidentiality was guaranteed throughout the entire process (Beauchamp & Childress, 2013). Children need information they can understand; they must know what they are agreeing to participate in. For this reason the children themselves assented (Ford et al., 2007; Neill, 2005) to participate, after receiving repeated age-appropriate child versions of study information, both written and verbal. The younger children received consent forms written in simpler language – and all participants personally signed informed consent-forms.

Parents first received written and oral information about the study from the hospital nurse or physician involved in the family’s care. Families were contacted only if the child together with the parents consented to participate. This allowed the decision about participation to be made in private, within the family. Regarding the principle non-maleficence it was important to be aware of the unequal power relationship between a child participant and an adult researcher (Kirk, 2007). Bearing this in mind the researcher tried to create a space which would enable the participants to speak openly. It was vital in Study II and III to emphasise that the child knew everything about their own life and I, as an adult, was interested in their experiences and that there were no wrong answers. The participants decided where they wanted to meet, it was important to be in a safe and secure place (Coad et al., 2015; MacDonald & Greggans, 2008; Scott, 2008). In Study II and III children were asked to take photographs, prior to our meeting, of important things in their everyday life. The photos were used to stimulate and facilitate the interview and hopefully make the child feel they were in charge of the conversation when they told me about their pictures. The photos were used exclusively for the interview and have not been kept, analysed or interpreted separately. Beazley et al. (2009) have described right of children to be properly researched, using methods that make it easy for them to express their views and experiences. Participants in Study II and III decided whether or not they wanted a parent nearby during the interviews. As participants, particularly in Study II and III, were from a relatively small pool of people, strategies to protect participant identification were important.

This is an area where it is difficult to gain access to participants. These families often carry an excessive burden of care involving a large number of care activities and consequently leisure time is very valuable. Children are a vulnerable group in themselves and children on HMV treatment may also have major disabilities which can make them even more vulnerable. There were long distances to travel in order to meet these families. Scheduled meetings were canceled several times because of various infections and illness – a common problem for children on HMV. However the ethical principle concerning justice and beneficence means that a group cannot be excluded from research because the data collection is challenging. The Swedish Research Council (2017) states; “it would be almost unethical to refrain from conducting research on factors that may improve people’s health and living conditions, eliminating prejudices or raising people’s awareness of how to make more use of their own resources”. These words are applicable in the context of this study.

Results

This section provides an overview and summative presentation, study by study. The results from the four studies are described separately as they contribute in diverse ways to meeting the overall aim by providing more detailed in-depth descriptions of the PCAs’ experiences (I), the children’s (II) and siblings’ experiences (III) and finally the parents’ experiences (IV).

Study I

The findings in Study I are presented in accord with categories identified in the data.

The category “being part of a complex work situation” described a work situation which required mental preparation and alertness to anything that might happen regarding the HMV user’s breathing during the PCA’s work shifts. The participants had to leave the HMV user undisturbed sometimes, but still have control over the ventilator all the time - a challenging balance between closeness and distance. The “multidimensional responsibility” described a responsibility for another person’s breathing which incorporated being responsible for another person’s life. The participants also emphasised to another important assignment; empowering the person with HMV to take more responsibility for their own life, to grow as a human being and live as independent as possible. The HMV user’s life was fragile, which introduced existential dialogues about life, death and grief. Gratitude was expressed for being given the possibility to share another person’s life. The relation between the PCA and the HMV user
was characterised by a physical closeness. In some cases were also a very special “close emotional relation” developed, which sometimes included family members. However, PCAs also experienced great challenges concerning suboptimal leadership and work organisation - which caused them worries and stress. They told about their frustration that no health-care person had the overall responsibility for the HMV user. They struggled for the right of the HMV users’ to get what they were entitled to. They described feeling of loneliness; in some respects they were a part of the family and in other respects they were not. They worked with advanced medical technology in “someone’s home”, with various alarms and technical equipment’s to get used to and feel comfortable with. There was also a risk of being confronted with duties they perceived as morally unacceptable which caused worries, indecision and feelings of being both exposed and alone. To “create boundaries in an environment with indistinct limits” became a challenge. When working as a PCA for a child, questions could arise regarding the respective responsibilities of the PCAs and the parents.

Study II
The findings in Study II revealed that everyday life on a ventilator could be described as a duality that included a strong life spirit but a simultaneous fragility in face of the outside world, like balancing on a tightrope. Children on HMV told about an everyday life that included experiences of not being sick anymore, and even having plans for a future which could include working as a steward in an aircraft, despite the wheelchair and ventilator. They dreamed about a future life chosen by themselves, that fitted their preferences. Their vital technology included information and communication technology (ICT) and medical technology, which together were the enablers that made involvement in a world outside the home possible. The HMV device itself was easy to carry with them, just like a computer bag, but nonetheless it was sometimes a challenge to use it if friends made inappropriate comments. They socialised with friends via social media and online computer games. Being able to handle a mobile phone or game control on one’s own, without help from PCAs or parents, was an important detail. A really good day was a day that involved fun activities outside the home; for example going to friends’ homes after school and participating in sporting activities. This however, required PCAs - which they could be denied – making it impossible to participate in outside activities. Activities that many young people take for granted were not self-evidently available to them. Being forced to have PCAs constantly close to them was not a walk in the park either. When the relationship between them did not work out well it was very taxing to be physically close for many hours, at home, to a PCA they did not like. They had to adapt to given conditions - which in turn could be the result of political decisions, the municipality’s assessment of assistance needs, at a period in life when liberty was important.

Their everyday life included joy and power besides an extraordinary fragility; a daily balancing act between being sick and being healthy. They told of a constant fight against upper respiratory tract infections. Their ability to breathe was affected by a slight cold, and pneumonia could require intensive care and several weeks in hospital. Infections left them tired and some days could be filled with fatigue and respiratory tracts that had a disturbing amount of mucus, which had to be constantly coughed up. They were not able to run with their friends, not able to keep up and play with friends who went outside the schoolyard. They had to plan activities in advance, to be prepared, when they were very reliant on battery charges and susceptible to the effects of power outages. But, nevertheless, they also experienced feelings of never being sick anymore.

Study III
The meaning of having a sibling with HMV was understood as a complex and profound intertwined bond that linked to the past, the present, and the future. The siblings’ stories reflect such character traits as maturity, empathy and knowledge but at the same time they narrate experiences of worrying about periods when the disease is exacerbate, concerns about parents’ anxieties, responsibility for the ill sibling, being forced to grow up fast, and having limited time and space with their parents. The intertwined bond had shaped the participants to become prepared to fight for diversity and defend their sibling’s right to be themselves.

The ventilator itself was seen as a detail that their sibling with major disabilities used; a detail that did not affect their lives. But there was one exception; if the sibling had a tracheotomy attentiveness was required from parents and PCAs in a way that also restricted the sibling’s everyday life. They were used to having PCAs in their home, the younger children spoke about PCAs as fun friends but for some it was impossible to feel comfortable having people in their home, especially at night. The older siblings described this physical closeness to people and equipment as something they wanted to distance themselves from. They created (or longed for) areas in the house where they could be alone and have a sanctuary, free from PCAs, wheelchairs and medical devices. The siblings handled the responsibility by trying to
stay as healthy as possible and were aware that what they did could affect the sibling. The older siblings had realised that they took responsibility to a greater extent than their peers and they learned early how to cope on their own when their parents were very busy. There were glimpses of sadness in their stories about what was never going to be – they were never going to jump on a trampoline together or just be together in pajamas at breakfast on weekend morning without PCAs being around. On the other hand, they were aware that everyday life would never work out if the PCAs were not nearby.

**Study IV**

In total 85 parents: 45 mothers and 40 fathers of 55 children with HMV responded to the questionnaires in Study IV. Both parents responded from 30 families, in the other cases (n=25), either the mother or father did so. The parents’ mean age was 41 years, the children’s mean age was 8.5 years, and the mean time on HMV treatment was six years and eight months. The largest group (n=31) had NIV treatment. The result show that parents of children on HMV reported low HRQoL and family functioning in comparison to previous research which addresses HRQoL among parents of children with long-term conditions. One in four parents reported moderate or severe clinical insomnia. Mothers’ reported clinical insomnia to a greater extent than fathers. However, no significant differences were found between mothers’ and fathers’ HRQoL, family functioning or sleep.

Parents of children with tracheotomy reported more impact on HRQoL than parents of children with NIV and CPAP. This seems understandable, since children with a tracheotomy require monitoring day and night and may also have complex disease conditions that involve multiple technologies. The child’s HMV mode and the parents’ sleep quality predicted the parents’ HRQoL. The regression model indicated that 45% of the variability in the dependent variable Parent HRQoL Summary Score was predicted by the child’s HMV mode and ISI reports. The child’s HMV mode and ISI total score were also found to predict the Family Functioning Summary Score, explaining 21% of the variance.

**Discussion**

**Methodological considerations**

**Overall design**

This thesis has integrated qualitative and quantitative components, comprising interviews and questionnaires, which together have contributed to producing a broader and deeper knowledge about experiences from the HMV context.

The methodological considerations for the qualitative approaches in Study I, II and III are discussed in terms of trustworthiness; an overarching concept which involves credibility, confirmability, dependability, and transferability (Lincoln & Guba, 1985). The methodological considerations for the quantitative approach in Study IV are discussed in terms of validity and reliability (Polit & Beck, 2010, 2012).

**Trustworthiness in qualitative research (I-III)**

**Credibility**

Credibility refers to confidence in the truth of the data and the interpretation of data (Polit & Beck, 2012). The objective in Studies I, II and III was to include participants with various experiences thus increasing the possibility of illuminating the research question from a variety of aspects (Graneheim & Lundman, 2004). There were approximately 300 children receiving HMV in Sweden 2019 (Swedevox, 2019). This is a small geographical dispersed group of children, and it has been a challenging process to include and arrange interviews times with the participants and their families.

The inclusion criteria in study I were not limited to being a PCA for a child, which somewhat facilitated the inclusion process. In Studies I-III a purposive sampling approach was used. In study I maximal variation was sought in the sample (Polit & Beck, 2010) regarding work experience. If study I had been done today, the focus would have been on PCAs working with children on HMV in order to optimise consistency with the other studies in the thesis. The initial invitation to eligible interviewees was made via the PCA’s employers. As a strategy this can be criticized since it afforded the employer an opportunity to choose the participants they preferred to participate and exclude critical voices. On the other hand, the participating PCAs narrated about both good and bad work experiences. In Study II one goal was to ensure maximum variation in terms of the participants’ age, sex, HMV support, underlying illnesses, respiratory clinic and as in Study III age, gender, and younger or older sibling. One
participant in Study II was 20 years old and therefore not a child. But this participant was able to share an entire life experience of growing up with HMV which was invaluable in achieving the aim of this study.

The participants in Studies II, III and IV were invited to participate by personnel in the healthcare system and probably had a well-functioning relationship with healthcare. This fact could have affected the goal of achieving a representative sample and we may have lost families with a complicated relationship with health care, which may be considered as a bias. We also failed to include non-Swedish speakers, which is a major problem as some voices are missing. Discussions have been held among the authors about the possibility of using an interpreter - failed to include non-Swedish speakers, which is a major problem as some voices are missing. Discussions have been held among the authors about the possibility of using an interpreter - but we did not manage to include participants where that need arose. The important question remains of how we can succeed in including children with HMV and their non-Swedish speaking families in future research?

One strength in this thesis is that the supervisor group represents an interdisciplinary research team from areas with different traditions, which gave rise to ongoing dynamic discussion about preunderstanding. Discussions among researchers from varying traditions are one way tackling complex healthcare issues (Katz et al., 2016). To ensure credibility categories and theme were discussed in the supervisor group during the analytical process until agreement was reached.

**Confirmability**

Confirmability refers to objectivity – that the findings represent the participants’ voices (Polit & Beck, 2012). All interviews - except two - in Study II and III took place in the participants' homes, with a flexible attitude to each participant’s individual wishes. The context is particularly important when interviewing children (Scott, 2008). There were continuous discussions about the advantages versus the disadvantages of having parents present during the interviews (Study II, III). Some research states that children should be given the opportunity to tell their story in privacy, without the influence of parents being present (Clark, 2011; Gardner & Randall, 2012). The intention was to interview the child in a separate place in the home. But this often proved impossible in practice when the presence of parents' with their unique understanding of their child, was required for the child's story to emerge (Gardner & Randall, 2012). This was especially true in Study II where the parents’ presence was sometimes essential as the child’s voice was so hard to hear. Nevertheless, all participants in Studies II and III were invited to decide whether or not they wanted a parent to be with them. The parent could sit beside them without actively taking part in the conversation, or supplement the child's story by asking questions of the child such as "Do you remember what you told me yesterday" or "How was it when we did it?". Careful data analysis ensured that it is the child's own voice that constitutes the result. All the authors read and interpreted the interview text and discussed themes, subthemes and quotations (Studies I-III). Also research seminars, discussions, and reflections in the doctoral student group have facilitated developing a critical stance to ensure rigor (Morse et al., 2002).

**Dependability**

Dependability refers to the stability of the data over time and conditions. It is closely related to credibility, as validity is to reliability in quantitative research (Lincoln & Guba, 1985; Polit & Beck, 2012). The author of this thesis (ÅI-S) conducted all the interviews and made all the transcriptions in Studies I, II and III – except eight of the interviews in Study I, which were conducted by the by two specialists in nursing in primary health-care who were enrolled on a masters’ programme, as described in the method section. There were prepared interview guides for all interviews but they were only used as a support if the conversation was difficult to bring forward. The interviews were discussed and evaluated in the supervisor group and also with experienced researchers and colleagues in the child interview area: the aim being to reflect and develop the interview technique.

**Transferability**

Transferability refers to the extent to which findings can be applied to other setting or groups. We tried to achieve this by providing detailed descriptive information about the research setting and study participants (Polit & Beck, 2012). The studies were carried out in a Swedish context regarding legislations and healthcare organisations. Research findings can hardly be generalized without considering national differences and variations in health-care provision. Differences in health-care system exist and may affect the possibility of generalising results. Nevertheless the results of this thesis may be valuable to those dealing with other groups of children (II), siblings (III) and parents (IV), and professionals (I) working with children with long-term illnesses. Polit and Beck (2010) argue that research evidence can be used only if it has some relevance to settings and people outside the contexts studied; without transferability there can be no evidence-based practice. However, the main work of transferability is done by readers and research consumers, if they consider the findings relevant to new situations; the readers transfer the results (Lincoln & Guba, 1985; Polit & Beck, 2010).
Validity and reliability in quantitative research (IV)

Study sample and sampling

Validity refers to the degree of inferences that can be made from the actual research (Polit and Beck, 2012). Study IV has a cross-sectional design, aiming to include as many parents as possible. The sample was framed by the Swedish National Quality Register for Oxygen and Home Respiratory Treatment (Swedevox), which is a strength in this study. The strength that a National Quality Register possesses was used when developing a context.

Respiratory clinics (n=17), identified via Swedevox (2019) were informed about the study and requested to ask parents about participating. There were approximately 300 children on HMV therapy in Sweden in 2019 (Swedevox, 2019) and a total of 88 parents were included in the study; 85 participants from 55 families responded to the questionnaires. The response rate must be regarded as reasonable. One strength regarding the external validity in Study IV is that parents from different geographical parts of Sweden responded to the questionnaires. This mirrors regional variations regarding type of residence, healthcare organisation and equipment support. There are variations between living in rural areas versus city areas in terms of preparation for emergency situations, accessibility to a hospital for example.

Paired data, from both mothers and fathers, were obtained from 30 families. Parents were consecutively invited to participate by healthcare professionals, which meant that we could not affect the inclusion process. There is a lack of information about how many parents declined to participate and how many agreed to participate but did not return the questionnaire, i.e. non-responders, which much be regarded as a major limitation. It is understandable that there might be problems returning a questionnaire that was handed out at the hospital, but there were also problems finding time for parents to complete the questionnaires while they were at the hospital. One explanation for why the response rate was not higher may be that parents in this context are presumably occupied with responsibility and care, and probably have a lot of paperwork to do connected with authorities, resulting in their lack of answer further questionnaires.

The measure

Validity in relation to questionnaires is the degree to which the instrument measures what it is supposed to measure and reliability refers to the consistency with which it measure the target feature (Polit & Beck, 2012). One strength of Study IV was the use of two validated questionnaires; the Family Impact Module and the ISI. The Family Impact Module has well-documented validity and reliability (Jastrowski Mano et al., 2011; Medrano et al., 2013; Panepinto et al., 2009; Scarpelli et al., 2008; Varni et al., 2004). It is translated into Swedish (Tiberg & Hallstrom, 2009) and used in Swedish research (Gårdling et al., 2018; Jonsson et al., 2015; Jönsson et al., 2016). ISI has a documented reliability and validity for detecting insomnia (Morin et al., 2011); it is also translated into Swedish and has been used in population studies both nationally (Jansson-Frojmark et al., 2016) and internationally (Morin et al., 2009). The cut-off point is a score that separates cases from non-cases (Polit & Beck, 2012a). ISI reports were dichotomized, <15 and ≥15, according to the defined cut-off values where ≥15 indicates clinically significant insomnia (Bastien et al., 2001).

There is no defined cut-off value in the Family Impact Module and we did not have a control group from a Swedish context, which must be regarded as a limitation. The results from Study IV are compared with previous research both national and international, focusing on HRQoL among parents of children with long-term conditions. It would however be helpful to have cut-off scores for this measure when clarifying the effect that a child’s long-term disease has on parents.

Parametric methods were used for statistical analyses after data had been tested for normality (Skewness and Kurtosis) and criteria were fulfilled. Comparisons of mean values between cohabiting parents were performed via using the paired t-test. The paired t-test was chosen because the couples were considered to be dependent on each other, i.e. paired sets of participants (Polit & Beck, 2012).

Regression analysis is used to make predictions (Polit & Beck, 2012) and linear multiple regression with backward selection was selected, after normality, linearity and homoscedasticity were controlled for (Tabachnick & Fidell, 2014). The aim was to explain how much of the variance in the dependent variable (parents’ HRQoL Summary Score and Family Functioning Summary Score) could be explained by the independent variable (parents’ gender, parents working as PCA or not, parents having paid work outside the home or not, HMV mode and ISI total score). A linear multiple regression also gives an indication of the relative contribution of each independent value (Pallant, 2013). When linear multiple regression with backward selection is used all predictors are entered into the model, the weakest predictor is removed and the regression is recalculated until only useful predictors remain in the model (Brace et al., 2013). Backward selection was chosen because no strong hypothesis was formulated in advance, due to the absence of earlier research within this context.
Following the suggestion of Tabachnick and Fidell (2014) about the ratio of predictor variables to total number of cases, a minimum of 90 cases was considered sufficient for the five independent variables. Since Study IV had 85 participants five independent variables were selected. A review of previous research identified five factors that could possibly influence parental HRQoL; gender (mother or father) (Nozoe et al., 2016), parents working as a PCA or not (Graham et al., 2016), parents having paid work outside the home or not (Graham et al., 2016), HMV mode (Cadart et al., 2018) and ISI total score (Meltzer & Booster, 2016). Future research could also identify variables not evaluated in this study, which may be predictors affecting parent reports. For example being single parent, education and income, housing situation and the child’s disease severity and age.

Discussion of the findings
The overall aim of this thesis was to explore experiences of living with HMV from the perspectives of children, siblings, parents and PCAs. The participants’ everyday lives can be described as a fragile balancing act between health and illness and hope and concerns in some ways resembling walking on a tightrope. One side of the tightrope was positive and future oriented and the other side was characterised by uncertainty and concerns for the future. This way of existing can be compared with Gadamer’s (1996) description of health as a kind of equilibrium condition; a rhythmic experience when life is in harmony and people find their own paths in tune with the world. This thesis makes new knowledge available about everyday life in the HMV context which could hopefully support health-care professionals in assisting these families to find their own paths to a condition of equilibrium.

Being a PCA
The PCAs in the study told of the gratitude they felt to have a job that gave them a chance to share another person’s life, but at the same time they also described facing great challenges. Less than optimal leadership and organisation of the work caused them worries and stress, and the PCAs often felt lonely at work. The relationship with the management is important for job satisfaction and a more optimal integration of home-care workers into the home-care team has been suggested (Ryvicker, 2018). In Sweden, in some cases, family members are employed as PCAs, often together with external PCAs (Dunér & Olin, 2018), which means that the child’s parent can be the PCA’s colleague from time to time. The person cared for can be the employer and the carer the employee, which also raises questions about the balance of power (Clevnert & Johansson, 2007; Olin & Dunér, 2016). This is an area which there is generally very limited knowledge, and within the HMV context in Sweden there is an absence of research.

When the PCAs were confident about the ventilator it was not usually a big problem, but when they did not completely understand the equipment it could cause them anxiety and stress. When working with a person with a tracheostomy the PCAs tried to prepare for emergency situations by thinking in pictures, for example; “What will I do if the cannula comes away when we are in the shopping mall”? It was invaluable to have access to a knowledgeable mentor if problems or questions about the device occurred. There were challenging ethically complicated life situations to handle, for example, young people with self-destructive ways of living. Lacking moral confidence, not feeling qualified and not knowing how to respond to a morally challenging event can cause moral distress. PCAs working with children and young people receiving HMV frequently carry out advanced duties (Ramsey et al., 2018) and can be expected to run into situations that may cause moral distress. Lack of ethical knowledge often brings with it a lack of a mutual moral vocabulary as colleagues. This can cause another type of powerlessness, where PCAs are less likely to participate in interprofessional moral dialogue, even though they experience the same ethically distressing situations as their colleagues (Fryer et al., 2016; Rodger et al., 2018). The PCAs spoke about a work situation where they knew that they were going to lose both a dear friend and simultaneously their employer. Long-standing and close relationships with families can contribute to moral distress. Caring for a very familiar dying person and coping with death can be emotionally and physically exhausting and morally challenging (Fryer et al., 2016).

Being a child with HMV
The children with HMV described feelings of never being sick anymore. They talked about a future-orientated everyday life where they had power and energy and they had plans for education, employment, and an independent life. But at the same time, everyday lives were characterised by fragility, struggles against upper respiratory tract infections and an environment that in some ways made them disabled. If the battery of the HMV device lost their charge or the powered wheelchair did not work, they lost their opportunities to participate in outside activities. The children exemplified a bad day as a day when the electric wheelchair did not work. Their technology was lived to the highest degree.
Adolescents and young adults with congenital heart disease have described their existence as living in two worlds; a duality of one world with sickness and one world with health – and trying to find a balance between these extremes (Berghammer, 2012). Young people diagnosed with multiple sclerosis (MS) have described how their appearance of normality was continually threatened by unpredictable symptoms and physical changes. Fluctuating periods of illness and wellness challenged participants’ identity construction and created difficulties in incorporating illness into their lives (Kirk & Hinton, 2019). The core concern across all issues, among children and young people with a long-term condition, has been described as a desire to be like their peers, to fit in and be accepted (Dellenmark-Blom et al., 2016; Lambert & Keogh, 2015). This is in line with the narratives in Study II, where the children found it difficult when they could not keep up with their friends and when everything they were supposed to do required preparation. Using a breathing mask could be okay, as long as no friend commented on it in an undesirable way. Social connectivity and being accepted by peers are important issues for all children including those with long-term conditions (Kirk, 2010; Lambert & Keogh, 2015).

Bury (1982) developed the concept disrupted biographies; a long-term illness is said to disturb and control a person’s life, to challenge bodily assumptions and functions that are taken for granted, leading to the rethinking of a person’s biography and self-concept. Taghizadeh Larsson and Jeppsson Grassman (2018) have argued for the use of the concept to understand a more complex set of experiences in connection with repeated transitions due to bodily and functional losses over the life span in long-term illness; expected unexpected and feared losses. The concept has been used to examine how children and adolescence with long-term conditions experience diverse and changing biographical experiences, ongoing transitions and disruptions while growing up into adolescence and adulthood (Bray et al., 2014; Monaghan & Gabe, 2015; Saunders, 2017).

**Being a sibling**

The siblings’ narratives about their everyday lives portrayed them as being mature, empathic and knowledgeable but simultaneously worried and concerned, taking a lot of responsibility, being forced to grow up quickly, and having limited time and space with their parents. The siblings in Study III handled the responsibility by trying to stay as healthy as possible and being aware that if they caught a cold - which was not that serious – their sibling on HMV could also become infected. There was always a risk of serious illness. Taking home a friend with a sore throat was not a suitable thing to do. The loss of companionship between the healthy and the ill sibling has been mentioned among children growing up with a brother or sister affected by a long-term illness (Knecht et al., 2015). A lot of lost opportunities, common in other siblings relationships, for example being able to play with their brother or sister has been described (Knecht et al., 2015). A longing to be able to play, a wish to be able to do exiting things like jumping on a trampoline together have also been described in this thesis. All siblings were used to PCAs being at home. Castor et al. (2018) have described that siblings felt acknowledged when homecare professionals had time to talk to them about pets or hairstyles in a nice, friendly way. In some cases they were satisfied when the homecare professionals merged into the home environment without interfering with them. This is in line with the siblings’ narratives in Study III, where the younger siblings regarded the PCAs as enjoyable extra friends at home. The older siblings described, from their perspective, how they created (or longed for) areas in the house where they could be alone and find sanctuary from PCAs, wheelchairs and medical devices.

**Being a parent**

Parents of children on HMV were invited to participate in this study through self-reported questionnaires where they reported low HRQoL and family functioning in comparison to previous research which addresses HRQoL among parents of children with long-term conditions. Family functioning is associated with many dimensions of child well-being (Leeman et al., 2016). It changes over time and may be worse during acute periods of stress when the family system is disrupted (Herzer et al., 2010). Psihogios et al. (2019) have shown a relation between family functioning and medical adherence and recommend routine assessment of family functioning aimed at identifying risks and protective factors regarding adherence. Longitudinal investigations of family functioning that evaluate families as they progress (e.g., to school entry and adolescence), may help to identify the periods of greatest challenges (McClellan & Cohen, 2007).

In a Swedish prevalence study (n=1128) (Mallon et al., 2014) reported 10.5% of the adult sample insomnia disorder. Since research emphasises (LeBlanc et al., 2007) that HRQoL impairment increase with insomnia severity and there are significant associations between insomnia and physical and psychiatric disorders (Mallon et al., 2014), it is important to highlight that 25 % of the parents in Study IV reported moderate or severe clinical insomnia.

**To watch and be vigilant**

To watch and be vigilant is a health-care concept which harmonises with the narratives in this thesis. There are narratives about an everyday life characterised by vigilance described as; not
daring to leave the adolescent on HMV at an overnight camp - even though there were staff there; not daring to leave the hospital when the child with HMV was critically ill, even though the sister had a cold; not daring to leave the young child with HMV and no own ability to breathe, without there being two adults at their side - every minute. Being vigilant means being prepared for the unexpected - for the worst imaginable thing to happen. To watch and be vigilant is both a state and an act; a basic human need that stems from love for those close to us (Fridh, 2012).

Two perspectives can be distinguished in the concept; the perspective of the one who is watching and the perspective of the one watched over. For those who are being watched, it can mean a sense of security; being able to relax in the assurance that someone trustworthy is present and protect against hazards (Fridh & Bergbom, 2006). At the same time it could be a challenge; the teenagers in Study II spoke about highly taxing situations when they were physically close for many hours, at home, to a PCA they did not like. But at the same time, they knew that PCAs were essential for them to be able to be a part of society. Relationships with others may increase as well as diminish the feeling of being vulnerable - hence, vulnerability may also be regarded as a relational phenomenon (Gjengedal et al., 2013). The PCAs in Study I told of a challenging balance between being close and simultaneously respecting privacy; to leave the HMV user undisturbed sometimes, but still have control of the ventilator. The participants in Study III talked of their worries about their sibling’s disease getting worse, worries about parents being worried and a sense of responsibility – in some way like keeping a watchful eye over the family. One in four parents in study IV reported moderate or severe clinical insomnia and low HRQoL and family functioning. Watching over a child with HMV involves an emotional commitment, which can mediate happiness and worries - as all parenting does – but here the situation can develop into a state where parents surrender to the situation and set their own needs aside. This can be related to Eriksson (1986) who perceives basic human care as ontological; it is both a gift and a burden, but it is something that makes us human. Løgstrup (1994) formulates this as the crucial definition of the ethical demand; silent but imperative in its claim that effort has to be made, flowing from life itself, which means that the lives of our fellow human beings are constantly placed in our hands.

Professional vigilance is the essence of caring in nursing (Meyer & Lavin, 2013). The new insights gained from this thesis also make it important to create knowledge about what vigilance can mean in the HMV context; both from the perspective of being vigilant as close relatives, but also from what it can be like to be watched.

Transition
The concept of transition and the transition theory seem suitable for application in the HMV context. The common properties of transitions are processes, directions, and changes in fundamental life patterns (Schumacher & Meleis, 1994). Transition conditions are circumstances that influence a person’s way through a transition; conditions that facilitate or hinder a movement toward a healthy transition. Conditions that may influence the quality of the transition experience and the consequences of transitions can be personal conditions (emotional and physical well-being, knowledge, preparation and expectations) community conditions (resources and knowledge in the community) societal conditions (attitudes). Successful transitions may mediate a subjective well-being, role mastery, and satisfying relationships (Im, 2014; Meleis et al., 2000; Schumacher & Meleis, 1994).

The everyday life with a child on HMV seems to be characterised by transitions. For example after spending a long period in hospital, careful planning takes place to move from hospital to home. The transition of infants with HMV from hospital to home is a complex process requiring extensive coordination between the hospital and the family (Benscoter et al., 2019; Breneol et al., 2017; Toly et al., 2019). The transition to a portable home ventilator is one example of a major step prior discharge from hospital to home for a child requiring HMV (Fierro & Panitch, 2019).

More children on HMV are growing into young adults today due to advances in medical treatment and technology (Abbott & Carpenter, 2014; Simonds, 2016; Yamaguchi et al., 2019). Children with HMV are a diverse group with combinations of disabilities needing unique support, belonging to numerous pediatric sub-specialties (Brookes, 2019). The transition from the pediatric care to the adult healthcare system may be challenging for both children and parents (Joly, 2015; Onofri et al., 2019). Transitions into adulthood have been recognised as a particularly sensitive and challenging time for young individuals with long-term childhood conditions and their families. It is an important developmental stage and failure to get through it successfully may have consequences for the whole family (Waldboth et al., 2016). Young people with complex medical conditions and their parents have described the transition process as being like falling off a cliff, including feelings of abandonment, loss and uncertainty. Research has highlighted the importance of implement a nursing care
coordinator to ensure successful transitions for children with complex medical requirements (Amin et al., 2017; Breneol et al., 2017; Brookes, 2019; Hewitt-Taylor, 2012; McCasland & Brooks, 2019; Sparud-Lundin et al., 2017).

There seems to be a gap between how young people and their parents experience the transition to adulthood and what they believe would have facilitated it (Joly, 2015). Young people probably want to be more independent, while at the same time their physical disability and dependence on their families support increases (Waldboth et al., 2016). Being a parent of an adolescent with cerebral palsy (CP) in transition has been described as living in a time of change, with sorrow and anger that originates in the first grief of becoming a parent of a child with a disability. They felt insecure about what support their adolescents would be able to get in the future (Björquist et al., 2016). Chronic sorrow has emerged as a concept within which caring for a child with long-term illness or disability can be understood. When parents become aware of the impact the illness or disability will have on the child's potential to live the life they want, intense feelings of loss might emerge (Björquist et al., 2016; Coughlin & Sethares, 2017). This has been reported to happen when the child grows and fails to meet expected age-related milestones, during periodic relapses in the illness with repeated hospitalisations, or when the medical demands increase.

The results from this thesis show that there are transitions in the HMV context including transitions in roles: the parental role often changed to include nursing dimensions; parents were working as PCAs for their child, closely involved in their child’s care. Parents sometimes ran their own assistance company; they were employers of PCAs - with everything involved in being an employer. The parenting may not decrease when the child grows up – it is probably a lifelong situation characterised by prolonged parental caregiving. Siblings told of being worried about their brother or sister with HMV, a worry that would probably also affect them in the future, when their parents grew older. Previous research has emphasised siblings’ worries about their ill sibling’s ability to manage their disease and take care for themselves in the future (Waldboth et al., 2016). This has been described as a “responsibility that never rests” (Jarling et al., 2019) – in another context – but one that has much in common with that covered in this thesis.

This thesis includes narratives from families including children with a large spectrum of medical diagnoses - children who have survived cancer, children with muscular diseases, genetic diseases and brain injury – children who had respiratory problems in common. Their main challenge in everyday life does not seem to be living with HMV – there seems to be something more essential to focus on when living with a long-term illness which also includes physical impairment. This was also discussed by the PCAs. Teenagers on HMV were just like other teenagers, sometimes sore and angry and refusing to listen. The PCAs wanted to help them to grow as people and could not let them to do whatever they liked just because they were on HMV treatment.

Conclusions

The results from this thesis contribute with new knowledge, from the insider perspective of the family concerning everyday life experiences when a child has HMV treatment. Voices that have not been heard before in research are presented. Hopefully the findings will be a contribution to health-care professionals’ understanding of how family’s adjustments to life are influenced by the child’s HMV treatment and, furthermore, what adjustments are needed on the part of the healthcare professionals.

In the care context of children on HMV is it important to acknowledge that those children may experience themselves as healthy and living an ordinary life, just like their healthy peers. The main challenges in everyday life for the children living with HMV do not seem to be the HMV treatment itself. The underlying long-term illness which also often includes physical impairment seems to be the main issue in the life situation. The results of this thesis indicate that everyday life in the HMV context is a fragile construction; in some respects it is like walking on a tightrope. The fragile construction also influences and affects the everyday lives of the families and PCAs. It is therefore important to acknowledge that each child with HMV is always in a close relationship with others but is simultaneously an individual person with their own perspective.

The concept of sustainability has a clear position within the HMV context. A society that embraces diversity is a rich society and, from a sustainability perspective, an inclusive society is vital. It is important to provide children on HMV with the prerequisites for an active and healthy life. In line with previous studies (Kirk & Hinton, 2019) school seems to have a significant function in supporting young people with HMV and encouraging them to achieve their academic potential when giving careers advice. It is important to support and understand siblings on their own terms and provide parents with the prerequisites for coping and keeping
healthy in their own life situation. This thesis shows that one in four parents reported moderate or severe clinical insomnia, and compared with earlier research which addresses HRQoL among parents of children with long-term conditions parents of children with HMV reported low HRQoL and family functioning. This is critical knowledge as research emphasises (Golfenshtein et al., 2016) that parenting stress increases negative outcomes for families where a child has a long-term illness. Finally but not least, it is important to assist PCAs so they can feel secure and supported at work.

Clinical implications

Care in the HMV context is often characterised by lifetime management. It is an extensive care relationship in which aspects of life other than medical problems are implicated, where the whole family is affected in one way or another. The concept of transition and the transition theory (Meleis et al., 2000; Schumacher & Meleis, 1994) seem to be useful in this context. Some aspects of family functioning may be more challenging at certain periods in life and at different developmental stages. The theory may be helpful in understanding the circumstances that influence a person’s pathway through a transition; conditions that facilitate or hinder a movement toward a healthy transition. One way to work with the concept could be to create possibilities for children with HMV and their families to report their well-being, HRQoL and family functioning in order to identify those periods when families may need extra support. High stress periods and their effect on parenting stress might need to be considered by health-care professionals.

Finally, it seems important to work in multi-professional teams around children and young people with HMV. Teams should include a key-person who carries the main coordinating responsibility who shares knowledge and gives the PCAs confidence and security. A deep understanding of vulnerability may also be helpful for health-care providers working in the HMV context. To understand the children, the families and the PCAs from their own perspectives seems to be crucial, and being sensitive to the vulnerability of the other could be a key to acting ethically.

Future research

The results from this thesis suggest further possible research:

- To examine everyday life experiences in families where a child receives HMV treatment and the family members speak Swedish as a second language with the aim of including diverse cultural perspectives.
- To examine the complexities of the transition process, from the health-care professionals’ perspective, when planning and carrying out the transfer of children and young people with HMV treatment from hospital to home.
- To perform a longitudinal study of children and young people on HMV and their families aimed at identifying certain periods and developmental stages where some aspects of family functioning may be demanding and complicated.
- To examine the social inclusion experiences of young people with HMV regarding employment and education, leisure time, and socialising with friends.
- To carry out an inventory of the state of knowledge among PCAs working with children on HMV and examine what support they request in order to be able to feel included, secure and safe at work.
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Mölndal, 13 November 2019
Åsa Israelsson-Skogsberg
Resultat

Delstudie I

De personliga assistenterna berättade om en komplex arbetssituation med otydliga gränser där de hanterade ett multidimensionellt ansvar. De balanserade kravet på att hela tiden ha kontroll på ventilatorn samtidigt som de försökte tillgodose en annan persons rätt till integritet. De efterfrågade mer utbildning och stöd, och berättade om svåra utmanande situationer där de känt sig utlämnade och ensamma. Samtidigt hade de genom sitt arbete utvecklat som människor, och i vissa fall grundlagt en ny syn på livet. Ofta uppstod en närhet till personen de arbetade hos, vilket kunde bli mycket svårt när personen ifråga försämrades eller avled.

Delstudie II


Delstudie III


Delstudie IV

Studie fyra visade att en av fyra föräldrar till ett barn med HMV rapporterade måttlig till svår klinisk sömnlöshet. I jämförelse med tidigare forskning som fokuserat HRQoL hos föräldrar till barn med andra långvariga sjukdomstillstånd rapporterade föräldrar till barn med HMV lägre HRQoL och familjefunktion. Resultatet visade inga signifikanta skillnader mellan mammors och pappors skattningar. Föräldrans HRQoL och familjens funktion predicerades av hur de skattade sin sömn samt vilket form av ventilatorstöd barnet hade.

Slutsats

Resultat visar att barn och ungdomar kan uppleva sitt liv som gott och innehållsrikt trots långvarig sjukdom och ventilatorbehandling. Syskonen beskrev exakta och detaljrika minnen från sjukhusvistelser och deras erfarenheter präglade dem senare i livet. Deras berättelser understryker hur viktigt det är att som syskon uppmärksammas och beredas eget utrymme. Föräldrar till barn med HMV skattade sin HRQoL, familjefunktion lägre än andra grupper av föräldrar till barn med långvariga sjukdomar. De personliga assistenternas berättelser handlade om att förbereda sig mentalt på att allt kunde hända och inte veta om support och stöd fanns att tillgå i svåra situationer.

Denna avhandling bidrar med ny kunskap om hur vardagslivet kan vara för familjer där ett barn har hemventilator, där hälsa och livskraft verkar samexistera med en skörhet och sårbarhet inför omvärlden. Med ökad kunskap är det möjligt att utveckla vårdstrategier som stödjer barn med HMV, deras familjer och personliga assistenter där utmaningar identifieras och förstås - och familjernas unika kunskaper och styrkor understöds och omfamnas.


Introduction

Medical advances and critical care have contributed to raising the numbers of technology-assisted people living in their own homes (1, 2). Increased attention to patients’ wishes has also played an important role in the development of high-tech home care (3). One such group comprises those using home mechanical ventilation (HMV). Thirteen women and two men participated; their working experience with HMV users ranged from one to 17 years (median 6 years). Data were subjected to qualitative content analysis in an inductive and interpretive manner. Findings: Five categories emerged from the data: Being part of a complex work situation; Taking on a multidimensional responsibility; Caring carried out in someone's home; Creating boundaries in an environment with indissoluble (with a facemask) (3). At the turn of the year leading to alveolar hypoventilation (CAH) (5, 6). Ventilatory support may be required either during sleep or over 24 hours (7) (invasively with tracheostomy) or noninvasively (with a facemask) (3). At the turn of the year 2011–2012 (8), there were 2300 HMV users registered in Sweden (24/100 000). Under Swedish legislation, people with various disabilities are given the opportunity to receive care from personal care assistants (PCA). The Swedish National Board of Health and Welfare (9) defined personal care assistant as: A personally tailored support that can be given to a person who, because of serious and permanent disabilities requires assistance with basic needs. Swedish disability policy is based on the principle of universal equality of dignity and rights. The purpose of personal care assistance is to promote equality in living conditions and full participation in society (10). The care recipients, in this case HMV users, are free to employ their own PCAs. Dependency on care may engender a feeling of inferiority and loss of independence (11) which is more easily coped with if the person can influence the organisation of their care (6, 12).

Leece and Peace (13) describe how disabled adults who employ their own PCAs have a greater sense of autonomy. Nevertheless, life on a ventilator can be a challenge. The ability to breathe is connected with life itself; it is an absolute physiological and existential need (2).

Conclusions: The participants felt very close to the person they worked with, both physically and emotionally. They had a great responsibility and therefore a commensurate need for support, guidance and a well-functioning organisation around the HMV user. There is international consensus that advanced home care will continue to expand and personal care assistance is key in this development. We suggest that one way to move forward for PCAs working with HMV users is to create multiprofessional teams led by a key-person who coordinates the individual needs. More research is needed within this area from a broad perspective including the HMV-assisted persons, relatives, personal care assistants and management organisations.

Keywords: home health care, ventilator care, home health aides, qualitative studies, assistive technology.

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Sweden

HMV users are forced to rely on both lifesaving devices and other human beings in order to sustain life and health. The ventilator compensates (14) for the missing function, that is, breathing. The experience of breathlessness has been described as an extreme and frightening symptom involving panic and fear of impending death (15–17). Research (14, 18–20) has pointed out that HMV is associated with a wide variety of ethical challenges and more knowledge is needed on how to integrate technology and the ethics of its use with care practices. Wang et al. (21) describe technology as complex and frightening for both families and healthcare professionals. Ahlström et al. (22) point out the challenge of having staff in the home day and night, with the accompanying loss of privacy, a HMV user can have up to twenty PCAs working in their home (23).

The organisng of PCAs’ responsibilities vary across countries but there is international consensus that more knowledge within this area is needed (18, 24–28). There are no formal competency requirements for PCAs working with HMV-assisted persons in Sweden (9) even though they are responsible for a life-supporting home ventilator and the HMV user’s everyday care and safety. The largest number of PCAs in Sweden are the municipalities but there are also private companies (29). Although personal care assistance is a relatively new area of research, it is known that difficulties may arise that do not occur in a similar way in other care occupations (25). It is an intimate work situation characterised by the PCA’s constant presence in the recipient’s life (30). When a professional enters another person’s private life, there may be difficulties establishing boundaries, in terms of work and leisure, friendship and a professional relationship (20). Tensions may arise because the recipient’s dwelling is both a home and a workplace (25).

Theoretical framework

Our theoretical framework is grounded in a caring science perspective, meaning that caring is regarded as unselfish and loving giving to the other (31, 32); it is characterized by responsibility and a wish to do good (33, 34). A person-oriented professionalism is about investing something of oneself in the meeting with others, about putting the other in the centre. It is not only about paid work, but also represents a fundamental human calling where engagement and closeness are central (35). Erikson (36) writes that human suffering can be alleviated by human caring and solitude; seemingly simple everyday activities can provide power and dignity. PCAs are in a position where they can ease suffering; they respond to the fact that someone depends on them (19).

Rationale

There is national and international consensus that the need for vital and competent home care will continue to increase in relation to changes in patient population and acute care (1, 19, 20, 26, 37). This situation constitutes a new and growing challenge for health care. People with impaired communication skills requiring HMV and form a small but growing group in our society (3, 21, 38–42), and HMV treatment is one type of advanced home care that demands new knowledge. MacIntyre (43) points out that there is a rapidly growing population of patients who have survived acute critical illness but are not no longer in a hospital but are receiving home ventilator treatment and HMV. The second group requires caregivers with unique skills encompassing critical care, rehabilitation competence and medical expertise. If HMV users are to feel that they are living a good life, it is essential that qualified PCAs work in their homes on a continuous basis (6). At the same time, research clearly shows that PCAs working with vulnerable HMV users in home settings are often left alone with responsibilities that far exceed their level of knowledge and training (20). Consequently, more knowledge is needed about the work situation of PCAs in relation to their experiences of caring for HMV-assisted people, and about their needs and requests in order to offer them adequate training and support in their work. In summary, there is little existing research in the area (5, 23, 27, 41, 44) which was the motivation behind this study. Therefore, the purpose of this study was to describe PCAs’ experiences of working with a ventilator-assisted person (adult or child) at home.

Methods

Design

Qualitative methodology focuses on understanding the human experience as it is lived. Data collection often takes place through a careful compilation and analysis of narrative and subjective materials (45). The decision to use interviews as the data source was a natural choice as we wanted to share each PCAs’ unique lived experiences (46). To ensure methodological coherence, Morse et al. (47) state that the research question must match the method which in turn requires the data analysis to be a cyclical procedure. Rigour has been considered throughout the process of analysis (48) from the choice of the most suitable data collection method that would answer the research question to carefully describing the analysis process (49, 50).
The data comprised a purposive sample and consists of tape-recorded semi-structured interviews with 15 PCAs. Seven interviews were collected by the first author (A.I.) and eight by two district nurses enrolled on a masters' programme. The interviews used a common thematic interview guide, which helped them to implement comparable interview situations.

The initial invitation to eligible interviewees was made via the PCA’s employers who received information about the study which they then passed on to the PCAs. Once a PCA had agreed to participate, they were contacted by the interviewer. Data collection took place from November 2010 to March 2011. A diary was used and notes were taken after each interview ensuring that impressions which emerged from the interview session would be remembered. The participants were all working as PCAs, supporting a child or an adult receiving HMV treatment to sustain health – and life itself (Table 1). No importance was attached to whether the HMV user was a child or an adult. Thirteen women and two men participated (Table 2). Some were employed by the HMV users and some by a private company or the municipality; some were accustomed to working in pairs.

Eleven interviews took place in the interviewee’s home, two in quiet room at a university, and two at the home of an HMV user. The interviews started with an introductory question: “Could you please tell me about your working experience with HMV-assisted people?” The tape-recorded interview guide (46), which helped them to implement comparable interview situations.

First the text was first transcribed verbatim, read and reread in order to gain familiarity with and a deeper understanding of the content; the text was then divided into parts containing similar content. A short word describing the content was written in the right-hand margin. Codes with similar meanings were grouped together as subcategories. By interpreting the data, subcategories containing similar events were identified and grouped into higher order categories, to describe and abstract the phenomenon of interest.

Findings

Five categories with subcategories emerged from the analysis (Table 3).

PCAs experiences of caring for people on HMV

Being part of a complex work situation

Work was not considered stressful, in the sense that they had to rush during their shift, but the working situation was demanding and energy-consuming in that they worked many hours and always had to be alert. The work demanded that they should be mentally prepared for anything to happen during their shift regarding the HMV user’s breathing. There was a balance to be kept between spatial closeness and distance in terms of leaving the HMV user undisturbed but still having complete control of the ventilator. The work could be very lonely:

When you work as a PCA you don’t have any work colleagues to talk to during the day. There isn’t any-one there like you are used to having. We often work long shifts and I miss having colleagues to talk to (1)

All the participants expressed a conviction that their work had great value. They were able to offer help and the HMV user and the family often expressed their appreciation.

Working as a PCA had given them a broader perspective on life and personal growth. They felt grateful to share another human being’s life experiences.

To have the chance to work in care, to get an understanding of what it means to be disabled. To realize that it is not over, there is a lot left to live for. I think that’s the big thing I’ve learned (8)

One participant expressed the opinion that personal care assistance is a “run-through” occupation, something that it is not over, there is a lot left to live for. (1)

Taking on a multidimensional responsibility

The responsibility comprises different aspects; it was multidimensional. The participants talked about the responsibility for another person’s breathing, actually for another person’s life with great respect; it was seen as heavy and took a long time to get used to. They had different ways of handling it but feeling secure in their knowledge about the medical devices was a prerequisite for taking on the responsibility. It was important that medical devices were reliable and easy to handle. Some of them tried to prepare themselves for emergency situations. Knowing exactly what to do and what prepared meant a lot. They had an “emergency bag” with essential equipment and they rehearsed imagined scenarios.

What do I do in an emergency situation? What do I do if the tracheal cannula comes away? I think like pictures in my head, what will I do? (12)

Another aspect of the multidimensional responsibility was connected with having a properly functioning organisation, to know that they had the support of their managers and peers when needed. This could concern technical questions about the ventilator and breathing problems, but also the feeling of having a peer behind you. The feeling of familiarity with the HMV-assisted person, knowing their preferences and values made the responsibility easier.

However, there were several obstacles to taking on the responsibility. Some participants spoke about a suboptimal organisation which caused them distress. They clearly remembered situations when they had problems and needed immediate help but received no support from within their own organisation and were forced to dial 112 and turn to an emergency ward. Such situations gave rise to a sense of vulnerability and made the responsibility difficult to handle.

Just think, if she dies on my shift [when the ventilator makes strange noises], I want us to go to the hospital but the HMV user won’t go. There is a conflict. But it’s a big responsibility for me to see to it that she gets air. (14)

Disputes with the family about how to perform certain tasks could also hinder taking responsibility. PCAs told...
about situations where they had been on a course and learned new skills for performing a special task but next of kin had a completely different opinion about the correct way to do things. In such situations, it took courage to oppose the next of kin.

Another dimension of the responsibility involved empowering the HMV-assisted person to take more personal responsibility for their life. This was regarded as a major goal. They wanted to help the HMV user to grow as a person and become more independent.

The interviewees talked of apartments to the HMV user. As easy to handle, but it could take years to get used to. Technology in someone's home was a multidimensional issue. Working with medical technology in a home environment was often a question of interpreting implicit wishes. The PCAs emphasised the fact that it is the HMV-assisted person who decides what to do and when to do it, but in some cases it was difficult for them considering the working tasks they were expected to do. Sometimes the PCAs felt anxious about certain tasks and there was a risk of being confronted with tasks they did not want to do, some of which they found morally unacceptable. This was a particular burden for those who worked alone. Drawing the boundaries then became a constant problem:

In the beginning she didn't want to have the [ventilator] mask on her face at all, she refused and resisted. But the parents wanted her to have it on. We have discussed this with the parents a lot, we can't, and we aren't allowed, but it's the setting of a boundary that we have been busy with all the time. Where is the limit for what is good for her? Should we force her? But at the same time she does not always realize herself what is for her own good. (12)

The participants who worked with a child sometimes experienced great difficulties in setting limits. Situations arose when the PCAs and the parents disagreed about child-rearing and what their respective duties were. The PCAs could see some tasks as clearly being the parents' duty, for example, helping the child to brush their teeth, while the parent saw it completely differently. They described how they did not want to argue or alienate the parents, nor to be perceived as a problem. Some of the PCAs were forbidden by the parents to talk about difficult situations with their colleagues, which were perceived as highly problematic:

When you work as a PCA in a family you can't talk all over the place, maybe not with the family and certainly not with your work colleagues. That can be hard. (2)

When the working situation was complicated, it was very important to have supervision from a mentor or manager to be able to set limits. The manager could also be a help in the dialogue between the PCA and the next of kin.

There was disparity in how the PCAs felt about the importance of maintaining a professional role rather than being a personal friend of the HMV user. Establishing good relations and getting to know the person were important. Some of the interviewees regarded finding where to draw the line between being professional and being a friend as being the basis of the profession.

When I go to my HMV user I don't feel that I am going to work. I'm going to visit a friend, that's how well it works between us. It's a great feeling but it's not always so appreciated by the employer. But it works brilliantly. (10)

Long working hours spent close to the HMV user and their next of kin could complicate setting limits for the PCA even if they were certain about the best way to proceed.

Being close to another's body and soul

Work was largely about being close to the other's body and their personal territory. To be physically close to each other, day and night;

As a PCA we sit close all night, perhaps 70 cm from the ventilator-assisted person and 40 cm from the ventilator. It's natural, if that person is supposed to live and feel well. (2)

The PCAs often worked with young people who wanted to explore life, go to concerts, movies, etc. In such circumstances, it was even more important to have constant physical contact with the HMV user as it was sometimes impossible to hear the ventilator's alarm. The PCA needed to have eye contact, see facial expressions or have a hand on the HMV-assisted person's shoulder throughout.

The character of the physical closeness could change according to the developments in the HMV-assisted person's health. In some cases, a change occurred because the HMV user's physical status improved, Today is the first day in two and a half years that I have left the mother alone [with her son]. She was alone with him for an hour. It's the first time since he was born. (1)

However, change could also occur because of gradual deterioration. The relationship then became even more based on physical closeness. In such situations, periodic conflicts inevitably arose between PCAs and users, and it became important to dare to face the conflicts. This was not always easy because the HMV user could also be the PCA's employer but it was important for the PCAs from several aspects:

She [the HMV user] is just like any other teenager. She is sore about something and won't listen. We want her to grow as a person so we can't let her do whatever she likes just because she's on a ventilator.

When you work as a PCA and you are part of a family, it is necessary to choose the boundaries within the family's sphere, that is a domain that seems to be especially reserved for someone else. Crossing boundaries could be complicated and the interviewees described how they constantly tried to detect what the family wanted them to do or be. The interviewees felt both personal and professional friendships with the HMV user, the next of kin, and the PCAs' employer. Their relationships were often complicated, as they worked in an environment that was neither their home nor a hospital. Consequently, they could not judge the user, nor a hospital, or a home environment which influenced how the PCAs wanted to work and the interviewees described how they constantly tried to detect what the family wanted them to do or be. The interviewees felt both personal and professional friendships with the HMV user, the next of kin, and the PCAs' employer. Their relationships were often complicated, as they worked in an environment that was neither their home nor a hospital. Consequently, they could not judge the user, nor a hospital, or a home environment which influenced the working environment was not always ergonomically organised. The interviewees talked of apartments.
respect for the lifestyle of the HMV user and the family. Lindahl et al. (31) write about professional friendship as a response in the HMV users and their families, where those who choose not to deliberately choose each other’s friendship. We believe that this concept can deepen the understanding and portrayal of the work performed by PCAs.

The interviewees, who worked with ventilator-assisted children, talked of working situations encompassing ethical dilemmas and uncertainty about boundaries. Previous studies (19, 21, 44, 52) show that the next of kin to a child with a ventilator and very watchful. Research (31) indicates that it may be difficult for PCAs to know how they should behave when people have differing ideas about their function and behaviour. However, little is written about personal care assistance for children.

The participants spoke of a responsibility to help the HMV user to grow as a person and maintain control over their own life. Earlier research (22, 54, 55) describes empowerment as a question of ethics and democracy and underlines the importance of supporting PCAs by providing them with ethical knowledge and supervision. Possessing this knowledge, the PCAs are capable of empowering disabled persons with self-esteem and dignity. These aspects can be compared to the participants in our study who made huge efforts to support the HMV users, not breaking free from the feeling of constraint. The results are in line with earlier research which claims that PCAs tried to make the most of their clients’ abilities to achieve the best possible quality of life (25, 56). When Lindahl and Lindblad (19) interviewed parents of ventilator-assisted children, the PCAs were described as being in a position to ease suffering. Fox (11) writes about two aspects of care, namely care as a gift and as a vigil; the concept that is a gift can create a positive loop with a reduced need for knowledge development, including a social and economic perspective, regarding the situation of PCAs and HMV users (54).

Methodological considerations

Inviting PCAs to participate in the study was not possible without help from their employers. This might have affected data. The first author, together with the second author, developed information about inclusion criteria was given to the employers in advance orally and in writing. The sample size is small but the participants represent what Politi and Beck (64) conceptualise as maximal variation and the PCAs mediated rich and diverse descriptions and meanings from various work-related situations. Within the framework of the study, we have not investigated whether there were any differences between the experiences of caring for an adult and those involving a child. This might be regarded as a weakness but further research could illuminate this situation. The authors have had discussions with peers and together reflected critically on the process throughout the analysis. Elo et al. (48) emphasise having a critical discussion about weaknesses and strengths in order to make the research process transparent and reproducible when using qualitative content analysis, thus supporting its trustworthiness.

Each qualitative research interview is unique, but a common thematic interview guide (46) to guide the interview in the same direction concerning the study aim was used. The interviewers were experienced nurses with further education, trained in scientific methodology. The participants were free to decide where the interviews would take place. The interviews were conducted by two experienced nurses who have been trained into English by a native speaker. Both authors are critical care nurses and have clinical experience of working with ventilator-assisted persons. NN is also an experienced researcher within the HMV area and in the use of qualitative methodologies.

Conclusion

Working as a PCA means experiencing physical and emotional challenges with regard to the care recipients. Work includes joy and pride but also feelings of loneliness and vulnerability; it also means carrying great responsibility involving major challenges. Today’s societal trend to move advanced care from the hospital to the low-palliative area will continue to expand. This change brings into focus issues concerning personal care assistance with regard to organisation and management and the field of knowledge and responsibilities of the PCAs. We suggest that one way to meet this challenge is to create multiprofessional teams around this kind of patient population, including a key-person who bears the main responsibility for coordinating all the issues and who shares knowledge and brings confidence to the PCAs. To conduct quality-assured advanced home care, easy access to professional support is required for the occupational group concerned. This applies to PCAs. More research is needed from a broad perspective including the HMV-assisted persons, relatives, PCAs and management organisations.

Acknowledgements

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Author contributions

Both authors have participated sufficiently in the work with manuscript to take responsibility for the content. A more substantial description is as follows: Asa Israelsson-Skogsberg is responsible for designing the study, recruitment of interviewees, the data collection process, analysis of data and the writing up of the manuscript. Berit Lindahl designed and planned the study, had access to the complete data set and contributed with intellectual input in the analysis process, in drafting the manuscript and revising it critically. Both authors approved the final manuscript.

Ethical approval

According to Swedish law (Codex, 2014), approval from an Ethics Committee is not necessary when a study concerns staff who is considered to consent to participation autonomously. The principals of the Helsinki Declaration (WHO, 2013) guided the contact process and consequently information was given about confidentiality and the possibility of withdrawing from the study without giving any reason. The participants received information and then gave their written consent.

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‘I’m almost never sick’: Everyday life experiences of children and young people with home mechanical ventilation

Ása Israelsson-Skogsberg, Lena Hedén, Berit Lindahl and Katja Laakso

Abstract

Developments in medical technology and treatment have increased the survival rates of children with serious illnesses or injuries, including those receiving home mechanical ventilation (HMV). Data were obtained through interviews with nine participants. The interviews were supported by photovoice methodology: photographs taken by the participants before or during the interviews were used to facilitate conversation. Interview data were analyzed using qualitative content analysis. The findings revealed that everyday life on a ventilator can be described as comparable to balancing on a tightrope. Various types of technology, both information and communication technology (ICT) and vital medical technology, enabled the participants to engage with the world around them. This study contributes knowledge about the experiences of children and young people living with home mechanical ventilation (HMV). Everyday life does not have to be spatially coherent—it may take place at hotels and airports all around the world. However, it does have a special life rhythm which includes breaks from daily routines (Ahrne, 1981).

Keywords

Adolescent, child, content analysis, home health care, mechanical ventilation, qualitative studies, young adult

Introduction

Developments in medical technology and treatment have increased the survival rates of children with serious illnesses or injuries, including those receiving home mechanical ventilation (HMV). This group is growing both in Sweden (Swedevox, 2015) and internationally (Rose et al., 2015; Wallis et al., 2011). The exact number of children receiving HMV therapy in Sweden is not known, but it has been estimated at about 300 (Swedevox, 2015).

The HMV can be delivered invasively through a tracheostomy tube or noninvasively through a face mask, and it may be required only part of the time, typically during sleep, or continuously for 24 hours a day (Chatwin et al., 2015). In terms of medical diagnoses, there are various reasons why children and young people may need HMV. The most common diagnoses requiring HMV include neuromuscular diseases, lung diseases, congenital central hypoventilation syndrome, and other congenital disabilities. However, a factor common to all of these diagnoses is alveolar hypoventilation (Edwards et al., 2010). Symptoms directly or indirectly associated with an impaired ability to breathe in children include disturbed sleep, attention problems, headaches, frequent infections, growth problems, and fatigue (Amaddeo et al., 2016).

Children receiving HMV may also have severe functional limitations such as difficulty walking, eating, swallowing, and, in some cases, speaking (Kirk, 2010). If so, they may need a range of assistive devices, such as gastrostomy tubes, oxygen supply devices, medical inhalers, and wheelchairs, a continual supply of equipment and frequent contacts with the health-care system (Heftner and Tsai, 2013). Because of their extensive care needs, some children with HMV require continuous care. In Sweden, children with HMV are supported by parental caregivers and personal care assistants (PCAs) for up to 24 hours a day. Prior research has shown that HMV users can develop very close relationships with their PCAs, based on emotional closeness and mutual respect (Israelsson-Skogsberg and Lindahl, 2016). The PCAs enable children and young people with HMV to participate in activities outside home and school. Indeed, research shows that, despite their severe physical limitations, many children with HMV consider themselves to have a good state of emotional and social well-being (Amaddeo et al., 2016; Verkaeren et al., 2015).

By definition, children with HMV live in their own homes (Carnevale et al., 2008). The increasing use of HMV represents a shift from health-care interventions in hospitals to professional support at home (Maddox and Pontin, 2013). ‘Home’ in this study is interpreted as a private space, a place for personal growth and everyday life, a place where to find rest and to live according to one’s own habits (Lindahl et al., 2011). ‘Everyday life’ refers to the circumstances where an individual experiences his or her daily needs for food, rest, and love. This term denotes an individual’s reality, and it includes the physical spaces, the society, and the culture in which that person spends time on a daily basis. Everyday life does not have to be spatially coherent—it may take place at hotels and airports all around the world. However, it does have a special life rhythm which also includes breaks from daily routines (Ahrne, 1981).

The general consensus today is that children and young people in need of ventilator support should not be institutionalized but should be able to use their ventilator equipment at home and bring it to school and other everyday settings (King, 2012). This requires advanced caring actions...
to be performed in the home, unique knowledge and training in those who provide the care and
treatment, and a transfer of financial resources from hospitals to homes (Boroughs, 2017; Sterni
et al., 2016). Recent research has identified several problems not only in preparing discharge from
hospitals but also in the provision of competent care in the home (McDougall et al., 2013). As a
result, children may have to stay in hospital for longer rather than moving home (Noyes, 2000), or
they may be exposed to insecure professional support at home (Boroughs, 2017). Moreover, their
parents may be forced to take on a heavy burden of providing care in the home for 24 hours a day
(Carnevale et al., 2008). This may cause one of the parents, often the mother, to stop working
outside the home (Olin and Dune´r, 2016). Using PCAs is one way to deal with these issues, but
they are costly and hence subject to much debate in Sweden (Government Offices of Sweden,
2016). Against this background, it is important to gain more knowledge about how children and
young people receiving HMV experience life. Listening to these children’s and young people’s
own voices is a prerequisite for gaining knowledge about how best to support this ‘new’ form of
childhood and parenthood (Graham, 2013) in order to offer those concerned good-quality care and
a lifestyle that they will appreciate. So far there has been only a small number of studies allowing
children and young people assisted by HMV to be heard (Falkson et al., 2017), which has been
described as a gap in the research literature (Earle et al., 2006). Hence, the aim of this study was to
explore everyday life experiences of children and young people living with HMV.

Design
Since the aim was to elicit the participants’ stories about their unique everyday life experiences,
interviews were considered appropriate (Kvale and Brinkman, 2014; Patton, 2015) combined with
photovoice methodology (Wang and Pies, 2004) to obtain data. However, given that some of the
participants had impaired communication skills and a weak voice owing to muscle disease and that
all of them suffered from shortness of breath, a carefully designed interview approach characterized
by sensitivity and flexibility was needed. Therefore, the participants were allowed to determine the
conditions of the interview, for example, they were given the choice of being interviewed with or
without parents present and whether they wished to paint drawings or talk about their photographs.

Participants and settings
The inclusion criteria were as follows: children and young people aged between 4 years and 21
years, who were supported by HMV or breathing assistance through a tracheostomy tube, and who
were at an adequate cognitive level and capable of telling their own story. Eligible participants
were recruited by nurses working at outpatient respiratory clinics at three hospitals in Sweden. A
purposive sample consisting of nine children and young people, five boys and four girls with a
median age of 11 years (Table 1), was selected with a view to ensuring maximum variation in terms
of age, sex, and type of assisted ventilation. All participants lived in their own home with their
families. Eight had PCAs. Three received parenteral nutrition and one received oxygen therapy.
The interviews were conducted between November 2015 and September 2016.

Ethical considerations
The study received prior approval from the Regional Ethical Review Board of Gothenburg,
Sweden (Ref. No. 354-15), and it was conducted in line with the World Medical Association’s

<table>
<thead>
<tr>
<th>Participant</th>
<th>Condition</th>
<th>Age</th>
<th>Ventilator regime</th>
</tr>
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<tbody>
<tr>
<td>Mio</td>
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<td>7</td>
<td>Face mask ventilator</td>
</tr>
<tr>
<td>Charlie</td>
<td>Lung condition</td>
<td>9</td>
<td>Face mask ventilator</td>
</tr>
<tr>
<td>Kari</td>
<td>Neuromuscular condition</td>
<td>10</td>
<td>Tracheostomy</td>
</tr>
<tr>
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<td>11</td>
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<tr>
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<tr>
<td>Loa</td>
<td>Neuromuscular condition</td>
<td>20</td>
<td>Tracheostomy</td>
</tr>
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</table>

Table 1. Characteristics of participants.

Declaration of Helsinki (WMA, 2013). The parents and children first received written and oral
information of an age-appropriate kind from the nurse at the hospital. Only when the parents and
the child had agreed to participate, were the families contacted by the first author. Written consent,
including permission for the photography session, was obtained.

Data collection
Semi-structured interviews were conducted in each family home by the first author. The partici-
pants were allowed to decide the setting for the interview, and they were told to choose a place
where they felt comfortable. The home is often perceived to be the child’s territory: a safe and
secure place (Coad et al., 2015). A thematic interview guide (Kvale and Brinkman, 2014) was
used.

As described by Wang and Pies (2004), photovoice methodology makes it possible to perceive
the world from a different viewpoint. Here, the participants were asked whether they wanted to
take pictures before our meeting, using their mobile phone cameras, of various aspects of their
home that they regarded as essential to everyday life. In some cases, photographs were taken
during the interview, and then, the whole photography session situation became a discussion
setting. The photos were used exclusively for the purpose of stimulating and facilitating the
interview; they have not been analyzed or interpreted separately.

Before each interview started, the participant was told that he or she was the expert on his or her
own situation and possessed unique knowledge and that there existed no right or wrong answers.
The aim was to initiate a conversation about everyday life. The interview started with the question,
‘Would you like to tell me who you are and what you like to do?’ There were large differences in
the narrated experiences of everyday life and the interviews were individually tailored to each
person’s wishes. We looked in photo albums, watched families’ private videos of memorable
situations, and read diaries from long hospital stays. Sometimes, the participants had taken photos
before the interview session, and in some cases, they borrowed the researcher’s digital camera and
took pictures during the course of the conversation.

Parents were present during six of the interviews, and a PCA was present during one. The
interviews were performed while life in general went on: siblings becoming upset and crying, dogs
harking, and visitors arriving. They lasted between 25 minutes and 90 minutes. The conversation
continued while we visited the participant’s room and looked at the breathing machine, photo
albums, and video recordings—even, in one case, while we participated in a football game. After
Future plans and dreams

PCAs provide essential support for an
Fighting against uncontrollable and
Computers, gaming and social media as
Being well prepared for various

The findings are presented in accordance with the two main themes identified: **Participation through technology** and **Balancing on a tightrope**. Table 2 summarizes the categories and subcategories below, illustrative quotations are given. These quotations have been translated from Swedish to English. The names used in them are fictitious and gender neutral which aims to eliminate the possibility of identification.

## Table 2. Presentation of subcategories, categories, and themes describing experiences of everyday life with HMV.

<table>
<thead>
<tr>
<th>Subcategory</th>
<th>Category</th>
<th>Theme</th>
</tr>
</thead>
<tbody>
<tr>
<td>Everyday life equals a well-planned structure</td>
<td>Everyday life composed by activities requiring extensive planning</td>
<td>Participation through technology</td>
</tr>
<tr>
<td>Being well prepared for various scenarios</td>
<td>Technology enabling an active and social life</td>
<td></td>
</tr>
<tr>
<td>Computers, gaming and social media as part of everyday life</td>
<td>Living between power and vulnerability</td>
<td></td>
</tr>
<tr>
<td>Technology of vital importance for life</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fighting against uncontrollable and invisible factors</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Illness impacting on body and life</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PCAs provide essential support for an independent and free life</td>
<td>Living between dependence and independence</td>
<td>Balancing on a tightrope</td>
</tr>
<tr>
<td>Creating meaningful relationships with others</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Future plans and dreams</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

HMV, home mechanical ventilation.

each interview, memos were written up about first impressions and contextual data regarding the interview situation. These memos served as a complement to the audio recordings in the analysis process (Morse et al., 2002), aiding the recall of the interview sessions.

## Analysis

The analysis took an inductive and interpretive approach to qualitative content analysis (Graneheim and Lundman, 2004). The first, preparatory, phase included immersion in the texts, photos, and memos. Then the interview text was transcribed verbatim. This was a slow, meticulous process. One issue that emerged only after we had listened to the recordings several times was that, in some cases, the parents and children had spoken simultaneously and finished each other’s sentences, because the child’s own voice was not strong enough or because the child did not have enough strength to conduct a long conversation. The different voices appearing in the transcribed sentences, because the child’s own voice was not strong enough or because the child did not have enough strength to conduct a long conversation. The different voices appearing in the transcribed sentences, because the child’s own voice was not strong enough or because the child did not have enough strength to conduct a long conversation. The different voices appearing in the transcribed sentences, because the child’s own voice was not strong enough or because the child did not have enough strength to conduct a long conversation. The different voices appearing in the transcribed sentences, because the child’s own voice was not strong enough or because the child did not have enough strength to conduct a long conversation.

## Findings

The findings are presented in accordance with the two main themes identified: **participation through technology and balancing on a tightrope**. Table 2 summarizes the categories and subcategories that describe our interpretations of the everyday life experiences narrated by the children and young people living with HMV. The participants’ narratives were rich and detailed—they had a great deal of knowledge to share about themselves and their illness, treatment, and medical technology. Their everyday lives did largely consist of challenges, but they also made it perfectly clear that this was their life and their way of living. In the descriptions of the various categories and subcategories below, illustrative quotations are given. These quotations have been translated from Swedish to English. The names used in them are fictitious and gender neutral which aims to eliminate the possibility of identification.

### Overall theme 1: Participation through technology

**Everyday life composed by activities requiring extensive planning.** To our participants, everyday life equaled a well-planned structure with a fair amount of important details. This could be associated with concrete instructions and directions, such as learning to be aware of one’s posture, always blowing one’s nose properly and performing various breath exercises several times a day. One young child explained that the purpose of this was to learn how to take care of one’s body and breathing. At school, it could be a matter of steering clear of anyone who had a cold and always using hand disinfectant; this is something that the younger children in particular were aware of. A young child’s photos of important things in everyday life featured a large hand-disinfectant container in the home.

All participants but one had PCAs assisting them with their physical needs at school, and they brought their technological devices back and forth from school every day. One young child brought additional food to school to meet the energy needs of breathing work. Furthermore, the teenagers living with HMV had one extra year in high school aiming to meet their need for individual adjustments.

Daily physical training, lasting for several hours, was a prerequisite for coping with everyday life. This workout was performed at home. All participants, even the youngest ones, initiated the workout themselves and assumed responsibility for it. As Kim explained, it was a natural daily routine:

> I get up at six or a quarter to six and get myself ready. Then I stand up reading for 45 minutes by a table, using the standing support. And of course I do the coughing machine before school, too.

The older participants experienced the ventilator as only a small part of their overall challenges in life. Still, as Alex pointed out, there was so much to do at school and yet managing one’s everyday life also required a great deal of work:

> After school and at weekends, it seems that everyone else has the possibility to do nothing, but I still have a packed schedule. Like, ‘No, I can’t do this, because now I have to go to hospital’.

In this context, the desire to spend time elsewhere than just at home or at school was important. To be able to be in an environment that was not well known, however, they had to be well prepared for various scenarios. For example, on the rare occasions when they slept over at their friends’ houses, they had to bring their medical devices and drugs along. One younger child had a special ‘sleeping over packing list’ to ensure that nothing important would be forgotten. Where a child had PCAs around the clock, they would sleep over with the child. One teenager described how he could be spontaneous in his activities and could go into town by car, although he always had to be accompanied by two PCAs when outside the home in case of problems with his tracheostomy.

**Technology enabling an active and social life.** The participants’ narratives underscored the impact of computers, gaming, and social media. In particular, for the older participants, gaming had...
immense significance and provided them with joy and happiness as well as feelings of sadness and loss, as described by Loa:

I’m happy when I play computer games, and I’m very sad when the Internet does not function optimally. It is a really bad day when that happens.

They socialized with friends via the computer and social media like other teenagers do. Being able to handle one’s mobile phone on one’s own, without help from PCAs or parents, was important to younger and older participants alike. Still, one example given by participants of all ages of a really boring day was a day spent ‘stuck with’ the computer when nothing else happened: a day of unwanted inaction at home. By contrast, a really good day was a day involving activities outside the home. One limiting factor in this context was not being assigned enough PCA hours for the outside home activities, in which case the participants were reduced to staying home with their computer.

Technology also meant access to streamed audio books and music. Listening to literature opened up new worlds and horizons which would be difficult to reach in real life. The relevance of technology to daily life went beyond the Internet. It obviously also included the participants’ respiratory devices and their powered wheelchairs, that is, technology of vital importance for life. When this technology did not work, they lost their opportunities to participate in activities outside home. In Kim’s words:

A not so good day is when my power wheelchair breaks down. I get upset. Angry.

The design of the respiratory device was characterized as vital: It had to be small, convenient, and easy to carry. The participants, both younger and older, described how they brought their ventilator back and forth to school, placed on the back of their wheelchair. The design issue was especially important in cases where everyday life could involve long flights to holidays abroad, overnight stays in sailing boats or in school gyms during sports tournaments as well as camping trips. To perform such activities, the support of the hospital’s technical department was of the utmost importance. For example, there might be a need for extra-long respiratory tubes and a rechargeable ventilator battery, as described by Charlie:

You have the world’s longest tube attached to your breathing machine. So that you can have the breathing machine at that end of the tent and sleep at this end of it, with the tube extending across the whole tent like an elephant’s trunk.

Some of the problems with the respiratory equipment described by the participants involved air leakage from the breathing mask and annoying alarms going off from the device. Finding face masks with optimal function and fit as their faces grew was a challenge, especially for teenagers. The younger children really appreciated their breathing machines: It was nice to rest with the ventilator and the machine helped them take deeper breaths. They were aware of its importance, knowing that without the ventilator their breathing would not work at all. As Charlie concluded:

Actually, the mask is quite nice to wear, there’s like cotton around it here.
The narratives were also about future plans and dreams. The participants’ ambitions and goals were to obtain an education and work as architects, journalists, flight attendants, game designers, and clowns. Their stories were about moving the boundaries of what was possible for them to do. With the support of their families and PCAs, some of the younger children managed to swim 200 meters while wearing a breathing mask or to sleep in a tent assisted by a ventilator. The importance of future plans and dreams was stressed by Tintin:

> Getting somewhere in life is important, getting ahead, school, continuing your studies.

It was also important for the participants to be able to liberate themselves, to be able to lead an adult life on their own without being tied to their parents. The older participants’ narratives were not so much about the wish to be healthy as about the wish to govern themselves, to have something that was private, that was theirs only. Tintin expressed this in the following way:

> If I could change only one thing, then I would like to live on my own, not sharing, having something that was only mine. Not having to go to my room to be alone.

For the younger children, the liberation process could involve participation in the decision-making process with regard to hospital treatment, as described by Alex:

> I feel sort of a little shy. And you’re supposed to tell [people at the hospital] things about yourself, and that can matter a lot, the decisions can be like really big. This is hard on you.

In brief, the participants’ dreams about the future mainly involved leading a life that they had chosen themselves, in the way that they preferred. In Taylor’s words, this might be something that allowed him to say:

> I’m an 18-year-old guy living here. On my own. With ten PCAs.

**Discussion**

Our findings are in concordance with those from research into mental illness in young people (Gowen et al., 2012), which has confirmed that online peer networks prevent social isolation by giving people their own voice and an opportunity for self-expression. Similarly, young people with cancer describe networking online as very valuable in their life in that it facilitates real-time communication and connection with peers and people in the same situation; social media have become young people’s preferred mode for communication and support (Gibson et al., 2016). The findings of the present study showed that it was important to have the ability to decide when to socialize via ICT and when to socialize through physical contact with peers. Truly having that choice was not self-evident to this group of children and young people.

It was striking that the participants expressed a great deal of power and energy while at the same time telling stories characterized by vulnerability—which we saw as similar to balancing on a tightrope. For example, they were exposed to the risk of being taken severely ill by viruses and bacteria that would be quite harmless to most people. Moreover, they could be denied PCAs; in which case, they would have to cut down on their participation in activities outside home. Many things that are self-evident in the lives of many young people were not self-evident to the participants in this study. For example, it was not self-evident that they could choose to accompany friends’ home after school, as this required extensive planning, nor that they could participate in sporting activities, as this required the involvement of PCAs. Gadamer (1996) characterized illness as a disturbance in one’s freedom, as a state that involves a kind of exclusion from life. In his opinion, health is ‘hidden’ in nature and does not merely constitute a state that one can feel in oneself. Rather, health is a state where one feels involved with friends and engaged in one’s everyday life. Gadamer’s way of describing health and illness is consistent with our participants’ narratives about their everyday lives. Hence, it is important to know what ventilator-assisted children and young people perceive as a good life and what resources they need to reach a state of health; this is in line with previous research in the field (Survay, 2008; Noyes, 2006).

The present study is based on children’s and young people’s own voices and testimonies. For this reason, there was an especially strong need for comprehensive and careful data collection and analysis. It was important to be aware of the unequal power relationship between the young participant and the adult researcher (Kirk, 2007) and to create a space which enabled and empowered the participants to speak. Since communicating with disabled children can be different from communicating with other children, research involving disabled children must be flexible and able to take into account each individual’s condition and requirements (Stalker and Connors, 2003). The presence of parents and PCAs was crucial to the conduct of the interviews; this is an experience shared by previous researchers in the HMV field (Kirk, 2010; Noyes, 2006). Some of the parents sat with their child whereas others stayed nearby while engaging in various tasks, going in and out of the conversation. The approach taken to ensure that the participants’ own voices would be heard was to treat the different voices (participant, parent, PCA) separately at first and then finally as one voice, so as to ensure credibility. The final narratives are always the participants’ own stories, even if sometimes introduced by the parent or the PCA.

The interview guide was a useful aid to keep the conversation going. However, the conversation flowed more freely when the interlocutors’ attention had a joint focus, such as a picture, a pet, or the ventilator.

An alternative study design that could have been used to obtain more in-depth data would be to include fewer participants and perform repeated interviews and observations. However, this would have placed a huge burden on the families concerned, and their schedules are very tight as it is. For findings to be trustworthy, they must be based on appropriate and rich data (Elo et al., 2014). The
credibility of the present study is enhanced by the fact that the participants represent different ages, sexes, types of ventilator support, underlying illnesses, and respiratory clinics. To further strengthen credibility, quotations are presented to illustrate the subcategories and categories identified. During the analytical process, tentative categories were discussed among all four authors until agreement was reached, so as to ensure credibility. When it comes to transferability, while qualitative research does not endeavor to generalize, the findings of the present study may be valuable with regard to other groups of children and young people with long-term illnesses as well.

Conclusion

This study contributes new knowledge about everyday life of children and young people living with HMV, generated through their own narratives. The stories they told reflected both power and an extraordinary fragility. What made participation in the outside world possible was technology in various guises as well as support from competent and sensible PCAs and parents. Hence, the starting point in planning and delivering good-quality care for these children and young people should be an individualized approach aiming to satisfy their needs and wishes, taking into account what each of them finds valuable in his or her life. This is linked to the moral duty of society to ensure that these children and their families are placed in a satisfactory situation after medical treatment in hospital is completed.

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References


Siblings’ Lived Experiences of Having a Brother or Sister With Home Mechanical Ventilation: A Phenomenological Hermeneutical Study

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Abstract
Over the past few decades, there has been an increase in the number of children receiving home mechanical ventilation (HMV), and in many ways, families have taken responsibility for the required advanced homecare, which has placed considerable time demands on the family unit. Little is known about the life situation of the siblings of HMV-assisted children; their own voices and an insider perspective are missing. The aim of this study was to illuminate the everyday life experiences of siblings of HMV-assisted children. Data were obtained via interviews with 10 siblings with a median age of 9 years. Data were analyzed using a phenomenological hermeneutical method inspired by the French philosopher Ricoeur. Four themes emerged, all of which suggest that a complex and profound intertwined sibling bond develops that links the past, present, and future. The findings of this study provide valuable information from an insider’s perspective about the meaning of having an HMV-assisted sibling. Family-focused care with particular attention and support for siblings of HMV-assisted children can encourage the development of internal strengths, self-confidence, and resilience.

Keywords
child, family, home mechanical ventilation, sibling relationships, family nursing

Sibling relationships can be the longest and closest relationships in one’s life (Knecht, Hellmers, & Metzing, 2015; A. Meltzer & Kramer, 2016). The relationship is ascribed rather than earned (Cicirelli, 1995) and contributes to the development of social and emotional skills and behavior (Whiteman, Beccera Bernard, & Jensen, 2011; Woodgate, Edwards, Ripat, Rempel, & Johnson, 2016). Pediatric disability and chronic illness often have a critically extensive effect on siblings’ well-being (Giallo, Roberts, Emerson, Wood, & Gavidia-Payne, 2014; Goeke & Ritchey, 2011; Hartling et al., 2014) with both negative emotional symptoms that may interfere with school functioning and positive outcomes, such as satisfactory self-esteem, social resilience, and maturity (Barr & McLeod, 2010; Gun, Lum, Wakefield, Nandakumar, & Fardell, 2017). As part of the first author’s doctoral research project which examines family life when a child has home mechanical ventilation (HMV) treatment, the current study focuses on providing unique knowledge about the life situation and relationships between the siblings. Previous research on this group is sparse.

Background
Research about the sibling experience when a child is ill (Gan et al., 2017) found regular visits to the ill sibling in hospital and increased responsibilities at home. Parents who tend to focus on the needs of the sick child may provide less emotional support and attention to healthy siblings—a family interactional pattern that may contribute to stress and anxiety. A systematic review (Limbers & Skipper, 2014) underscored that having a sibling with a more severe disease may increase the risk of the healthy sibling’s health-related quality of life (HRQOL) outcomes. Studies of healthy siblings who have brothers or sisters diagnosed with cancer (Jenholt Nolbris, Enskär, & Hellstrom, 2014; Nolbris, Enskär, & Hellstrom, 2007) reported that everyday life varied between joy, worries, and feelings of an existential nature. A responsibility to protect and advocate for one’s sibling outside the family was...
reported (Nolbris et al., 2007). Worries about the ill sibling’s ability to take care of themselves in the future were also found (Waldboth, Patch, Mahrer-Imhof, & Metcalfe, 2016).

In recent years, there has been an increase in children receiving HMV (Amin et al., 2017; Fine-Goulden, Ray, & Brierley, 2015; Rose et al., 2015; Wallis, Paton, Beaton, & Jardine, 2011) in Sweden (Swedevox, 2015) and throughout high-income countries (Amin et al., 2014; Fine-Goulden et al., 2015; McDougall, Anderley, Wensley, & Seear, 2013). The evolution of the HMV has taken place over the past 30 years (Amirnovin, Aghamohammadi, Riley, Woo, & Del Castillo, 2018), and a shift in the health care of HMV-assisted children from hospital to home-based care has occurred (Castro-Codesal et al., 2017; Chatwin, Tan, Bush, Rosenthal, & Simonds, 2015). HMV-assisted children are a diverse and unique group with complex health care needs, having severe conditions that may result in medical frailty and functional limitations (Edwards, Morris, Nelson, Panitch, & Miller, 2017). A wide variety of unique and complex care is given outside the hospital (Sterni et al., 2016) which, in turn, affects relationships within families (Gan et al., 2017; Woodgate et al., 2016). Managing a long-term disease where the whole family is involved (Cadart et al., 2018; Falkson, Knecht, Hellmers, & Metzing, 2017) and being faced with a unique set of challenges has been described as a family affair (Knaff et al., 2013; Knecht et al., 2015; Wright & Leahey, 2013). Caring for an HMV-assisted child has also been described as burdensome (Keilty, Nicholas, & Selkirk, 2017; L. J. Meltzer, Sanchez-Ortuno, Edinger, & Avis, 2015).

According to Swedish legislation (Swedish National Board of Health and Welfare, 2007), HMV-assisted children are entitled to personal care assistants (PCAs) for up to 24 hr a day. PCAs, because of their continued presence in the home, may be regarded as being a part of the family unit (Israelsson-Skogsberg & Lindahl, 2017). Research has underscored parents’ attempt to normalize family life (Toly, Blanchette, Sikorski, Musil, & Al-Hamed, 2017), but the required attention and numerous tasks required in caring for an HMV-assisted child may detract from other family responsibilities, particularly the well-being of siblings. Routines and planning around the clock for the HMV-assisted child influence the well sibling’s everyday life (Lindahl & Lindblad, 2011). Carnevale, Alexander, Davis, Rennick, and Troini (2006) reported that the siblings of HMV-assisted children experienced imbalance in parental attention and feelings of not being loved as much as the sibling.

Changing geographies of care, that is, from hospital to home, means families have taken on responsibility for the required advanced homecare, which has placed considerable time demands on these families (Castro-Codesal et al., 2017; Heaton, Noyes, Slopér, & Shah, 2005; Knecht et al., 2015; Yotani et al., 2014). Research with HMV-assisted children has found that these children depict their lives as good and valuable (Israelsson-Skogsberg, Hedén, Lindahl, & Laakso, 2018), but little is known about the siblings of HMV-assisted children. Many studies are based on parents’ proxy perspective, which cannot substitute for the siblings’ own voice and insider perspective (Knecht et al., 2015). We argue, along with other researchers (Gan et al., 2017; Toly et al., 2017), that data on the well-being of siblings, together with data from the HMV-assisted child, and from the parents, would provide rich and important knowledge about how these families manage their daily lives.

Listening to the voices of the siblings of HMV-assisted children is important. Therefore, the current study focuses on siblings to capture their own perspectives and experiences of everyday life. The aim of the present study was to illuminate the everyday life experiences of the siblings of HMV-assisted children.

Method

Methodology

The current study used a phenomenological hermeneutical method inspired by the French philosopher Ricoeur (1976) and later developed and described by Lindseth and Norberg (2004). Using a lifeworld perspective, the method aims to describe, gain insight, and mediate a deep understanding about what the life situation might be for another person; this method has been used in earlier HMV research (Dreyer, Steffensen, & Pedersen, 2010; Lindahl & Lindblad, 2013).

Participants and Settings

The criteria for inclusion included being a child or adolescent between the ages of 4 and 18 years and having an HMV-assisted sibling. Four nurses and one physician working at outpatient children respiratory clinics at three hospitals in Sweden recruited eligible participants. A purposive sample consisting of 10 siblings, four boys and six girls; three older, six younger, and one twin sibling with a median age of 9 years (see Table 1) were selected to ensure maximum variation in terms of age, gender, and younger or older sibling. All siblings’ parents lived together as couples; that is, there were no single parents.

All HMV-assisted siblings had diverse long-term medical diseases with extensive functional disabilities and a need for comprehensive support in their daily lives. All participants were used to having PCAs at home; five
HMV-assisted children had PCAs at home around the clock. Four HMV-assisted children received HMV around the clock, with six receiving it during periods of daytime rest and when sleeping. Eight had been using an HMV throughout their entire lives.

**Data Collection**

Data were collected via audio-recorded narrative interviews (Kvale & Brinkman, 2014; Mishler, 1986). These interviews were conducted by the first author between November 2015 and May 2018. The length of the interviews varied from 45 to 70 min. The siblings of HMV-assisted children were asked to take photographs with their mobile cameras prior to our meeting, capturing objects they thought were important in their everyday lives, which ranged from domestic animals and best friends to instruments. The photographs were intended to facilitate a conversation and were used only during the interviews; they have not been interpreted or analyzed separately. All interviews were flexibly designed (Spratling, 2013) according to each participant's individual preferences about where and when to meet, considering the studied families often have very tight schedules. Mostly, the interview took place in the family's home, a familiar and secure place, but in some cases, it took place at a café or an outpatient clinic. All the siblings decided whether to have a parent present or not. The older participants chose not to have a parent present, whereas the younger ones wanted to have a parent beside or nearby. After the introductions and during the interviews, the younger children often choose to draw a picture, present a favorite toy, or talk about their photos. This seemed to be a suitable way to enhance the child's level of comfort and establish a relationship between the child and researcher to better facilitate the conversation.

The open introductory question aimed to empower each child to talk: “Please tell me who you are and what you like to do.” This led to a dialogue about everyday subjects, such as the name of a domestic animal, a favorite football team, or life at school. When the sibling seemed to be comfortable with the situation, questions that aimed to evolve stories about their everyday life experiences as a sibling of an HMV-assisted child were asked as concretely as possible. For example, “Can you please tell me about a moment when you felt worries or happiness about your sibling.” Questions that clarified and explored their narratives were asked, “How did that make you feel?” The narratives of the younger siblings were very concrete and could consist of distinct memories of places, toys, and situations they had experienced, whereas the older participants narratives included abstract phenomena such as emotions and empathy.

**Data Analysis**

The interviews were transcribed verbatim and read several times, a naïve reading that aimed to grasp a first understanding of the meanings as a whole. Obtaining a naïve understanding is a starting point for the second steps of interpretation: a distancing and explanatory thematic structural analysis and the methodical instance of interpretation aiming to identify and formulate themes. Ricoeur (1976) considered a structural analysis as a necessary and fruitful stage in the critical interpretation process between obtaining first a naïve understanding and later a comprehensive whole with the end goal being to deepen the understanding and explain what the text discusses. Meaning units concerning siblings’ experiences of having an HMV-assisted sibling were identified, condensed, and abstracted (Table 2).

All condensed meaning units were examined for their differences and similarities. Narratives describing similar meanings were clustered and subthemes and themes were created (Table 3).

The third and last step of the analysis was to formulate a comprehensive understanding, the interpreted whole of the phenomena, where we considered all the parts of the analysis process and presented them in a new whole. We read the naïve understanding and the themes from the structural analysis and then formulated a new whole. We carried out the interpretation based on our critically reviewed preunderstanding and discussions with each other. This process entailed critical reflection on the results in relation to the relevant literature (Lindseth & Norberg, 2004). Our intention was to reach a deep
Table 2. Example of the Analytic Steps in the Structural Analysis.

<table>
<thead>
<tr>
<th>Meaning unit</th>
<th>Condensed meaning unit</th>
<th>Abstracted meaning unit</th>
<th>Subtheme</th>
<th>Theme</th>
</tr>
</thead>
<tbody>
<tr>
<td>I can talk a lot with him. We have a very good relationship with each other; we are probably quite the same and have very similar opinions. I say a lot of things to my brother, as I may not say to mom and dad.</td>
<td>I talk a lot with him. We have a very good relationship, are quite the same and have very similar opinions. I say a lot of to my brother, as I do not say to mom and dad.</td>
<td>A strong relationship with my brother</td>
<td>Living in a unique family circle</td>
<td>A strong relationship</td>
</tr>
</tbody>
</table>

Table 3. Structural Analysis.

<table>
<thead>
<tr>
<th>Subthemes</th>
<th>Main themes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Having strong relationships</td>
<td>Living in a unique family circle</td>
</tr>
<tr>
<td>Having grandparents nearby</td>
<td></td>
</tr>
<tr>
<td>Having personal care assistants in the family</td>
<td></td>
</tr>
<tr>
<td>Living an ordinary everyday life</td>
<td></td>
</tr>
<tr>
<td>Missing the life on the top floor</td>
<td></td>
</tr>
<tr>
<td>Being afraid to stay on the top floor</td>
<td></td>
</tr>
<tr>
<td>Having a private zone</td>
<td></td>
</tr>
<tr>
<td>Feelings of worries and guilty conscience</td>
<td>Carrying conflicting emotions</td>
</tr>
<tr>
<td>Feelings of indignation and anger but simultaneously comprehension</td>
<td></td>
</tr>
<tr>
<td>Feelings of disappointment, sadness, and grief but simultaneously joy and pride</td>
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<tr>
<td>To be a part of each other</td>
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<td>To compensate a lacking physical ability</td>
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<tr>
<td>To host an openness and acceptance to a person or to a life that might be a little different</td>
<td>Being in a intertwined belonging</td>
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<td>To become be used to (but still not)</td>
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overall understanding of the phenomenon in relation to our aim, the naïve reading, and the structural analysis.

All authors read the interview text material and were involved in the analysis process. There has been a continually ongoing reflexive and critical dialogue within the research group where all three steps in the analysis process were considered until an agreement had been reached. Rigor and reflexivity were also strengthened via an audit trail (Morse, Barrett, Mayan, Olson, & Spiers, 2002), a diary where memos that emerged from the interview session were written.

Ethical Considerations

This study was conducted to comply with the World Medical Association’s Declaration of Helsinki (WMA, 2013) and received prior approval from the Regional Ethical Review Board (Ref. No. 354-15). The parents first received written and oral information from the hospital nurse or physician involved in the families’ care. Families were contacted only if the sibling of an HMV-assisted child agreed to participate, which enabled a decision about participation to be made privately within the family. An age-appropriate written and verbal study information was given to the participants, stating that withdrawal from the study was possible at any time and confidentiality would be guaranteed throughout the entire process (Beauchamp & Childress, 2013). Permission for the photography session, including written consent, was obtained from the participants and parents. The participants in this study were from a small population, hence easy to identify. A careful strategy has been taken to reduce the possibility of identification, including meticulous selection of anecdotal examples and excluding specific information.

Results

The findings are presented as a naïve understanding, a structural analysis resulting in four themes and, finally, a comprehensive understanding.

Naïve Understanding

A naïve understanding is a first grasp of the whole regarding the message in the text, written in an everyday language (Lindseth & Norberg, 2004). Our naïve understanding is expressed as follows.

Having an HMV-assisted sibling is a challenge but at the same time a relationship that provides an infinite amount of joy. Being a sibling of an HMV-assisted child is natural and a taken-for-granted existence that might be
a little different from others, but not strange. It is experienced as gratitude for a unique family life situation that has shaped your developing identity. Having a sibling that is “different” means feeling a little different yourself. However, you also have the opportunity to develop values and skills that others in your age may not have. The relationship with your sibling does not include many conflicts, but there is an anxiety about what happens if the HMV-assisted sibling gets sick. Parents’ time and energy can be greatly absorbed by the sibling’s needs, but sometimes, grandparents close to the family serve as an additional asset. Being admitted to the hospital with your family for a very long time means missing this caring environment when you return home. Having a private space where you and your friends can be alone, a sanctuary without breathing machines, wheelchairs, and PCAs becomes important. It sometimes includes protecting your HMV-assisted sibling from an external threat, for example, schoolmates who utter inappropriate statements. But simultaneously there is a sense that you and your sibling are protecting each other; it is a natural commitment to each other.

A Structural and Thematic Analysis

The naive understanding guides the next step of structural analysis which explains the text and deepens understanding of what the text offers about the meaning of being in the world as a sibling of an HMV-assisted child. The structural analysis revealed four themes: living in a unique family circle, living on the top floor, carrying conflicting emotions, and being in an intertwined belonging. The subthemes are identified below.

Living in a unique family circle. A child undergoing HMV treatment requires a great deal of support and assistance, which often influences the family’s way of living. The family life means being different from other families; in some sense, the HMV-assisted sibling is like a sun, where the family members form planets circulating around the sun. Strong relationships emerge, where the HMV-assisted sibling becomes a close friend with whom to share values and who is perceived as a soulmate. The sibling relationship is characterized by consideration without conflicts: “Usually no one is arguing with him, just because it’s not really the same thing to argue with him as with the others.” Feelings of pride and gratitude about being each other’s sibling are experienced: “It is fun to be X’s sister. The best thing about my sister is that she’s my sister.” However, at the same time, grief about the illness and breathing supports that limit the ability to do activities together with the family appear: “If she did not have such a disease, she could have moved normally, and I could have done so many more things with her.”

Having grandparents who live nearby and are close to the family becomes very important. Grandparents sometimes act as PCAs and become an invaluable resource and security when a crisis occurs:

When my sister was very sick, we had to be in hospital all the time. For a year. Sometimes, I had to go home to grandmother and grandfather, but usually grandmother and grandfather lived with us in a big hospital apartment.

Grandparents serve as a sanctuary where the sibling can choose to stay instead of being in hospital. By having them so close, it becomes possible to choose whether to stay at the hospital or not.

Having PCAs in the family means always being at least one person extra. Some siblings experience this situation as impossible to ever get used to and others as something self-evident and joyful, like enjoyable extra friends at home: “It is pretty fun with my sibling; then, you will meet all the funny assistants.”

Living on the top floor. Having an HMV-assisted sibling was experienced as living on the top floor. Sometimes, the HMV-assisted child was critically ill and had to stay in the hospital for a long time. For the sibling, living on the top floor involved experiences of residing in a house near the hospital. In Sweden, these units are placed on the highest level in the building, where families with very sick children can stay for a long time. It is a physical place, literally on the top floor, but it was also a metaphor that life on the top floor sometimes meant having a privileged life. This life entails having a little bit of extra of everything such as attention, ice cream, and joyful activities in the play therapy while the HMV-assisted sibling was being cared for in the hospital.

Staying at the family houses’ top floor meant for some siblings that life just went on as it always had and some siblings actually missed living on the top floor when they were back home again: “It was really fun there, I even miss that hospital.” It also meant being involved in various activities that one did not do at home. Having a place where one meets new friends and other siblings in the same situation, which is valuable, especially when there was no possibility to meet one’s friends at home for a very long period of time. The concept of home had several meanings; it could indicate the top floor of the family house near the hospital or their real home, without any obvious preference.

Living on the top floor entailed having relationships with friendly hospital staff members: “They were very nice in the ICU; they were the kindest I have ever met. They made a chocolate cake for me on my birthday.” Memories from the time spent on the top floor include funny furniture, a balloon with a
cat, and an exact memory about where Daddy stood when having fun in the playground. These memories appear as solid and clear, even if a long time had passed. The hospital room or the siblings’ illness is not equally well collected, but memories from the play therapy and all funny things that happened there are intensely sharp: “I think more about when we were playing.”

Also feelings of being afraid to stay on the top floor is something siblings had to cope with when there were feelings of uncertainty about how ill the sibling was: “I do not know how serious it can be when she is sick. When she has surgery, it may go wrong, and then, things may happen and so. And then you get worried.”

From a literal standpoint, staying or having the possibility to stay on the top floor (i.e., a domestic area of a more private nature) when at home held a great value because this was a private zone with space and possibilities to be with friends, with no PCAs, breathing machines, or wheelchairs nearby. Friends could visit here, even with a sore throat, meaning not as much consideration and caution was necessary.

Carrying conflicting emotions. Having an HMV-assisted brother or sister meant carrying conflicting emotions, such as feelings of worry about both sibling and parents; for instance, this might include experiencing feelings of fear about frightening scenarios: “You can see that they get worried when she is ill. They get terrified; they do not want anything to happen. I am worried that they will become worried. It is a vicious circle.” Emotions of a guilty conscience about not spending enough time together with one’s sibling sometimes arose, which was especially noticeable when the sibling becomes seriously ill. These conflicting emotions inside also included feelings of indignation when unknown people disrespectfully stared at the HMV-assisted sibling:

There are quite a few who look very much at those with disabilities, who stay and watch. I just do not understand why because they are people, just like the rest of us. Why should they look at them extra much? It’s like going around and checking someone because he has brown hair and staring at that person just for it. That’s quite strange.

Anger was another conflicting emotion experienced in situations where classmates talked inappropriately about an HMV-assisted sibling. However, simultaneously, the sibling had the ability to realize that it might be hard for outsiders to understand their family’s special situation. Another example was wishing and longing to do ordinary things with one’s sibling, such as running and jumping on the trampoline together, even if you knew that it is impossible. This gave rise to a sort of disappointment when seeing others’ siblings

develop in a way where physical play together was possible: “If he did not have his tracheotomy, he would have been able to run now. I know one who was born after, and he can run now.” Feelings of sadness that the sibling’s life was like this would sometimes arise. However, simultaneously, joy and pride of being the only person in the whole world who could make a critical ill sibling laugh, even in the hospital, emerged.

Being in an intertwined belonging. Being in an intertwined belonging was like being a part of each other, which involves both physical and psychological dimensions. One participant provided this explanation:

We share a room and sleep together. The breathing machine is right next to me. At first, it was a little bit awkward because it sounded all the time. But now, when he is not there, it feels very weird sleeping because everything feels enormously silent. It feels better when he is here and I can hear him, and if he sounds strange when he is asleep, I tell Daddy.

To be a part of each other also meant a constant awareness that what one does affects the other; for instance, being without a cap at the schoolyard can cause one sibling a cold, and then, the HMV-assisted sibling may catch a cold. This almost always causes a respiratory infection, which frequently leads to a long hospital stay.

Having a sibling with a tracheostomy meant always having one or two PCAs at home, all hours of the day. But having a sibling with noninvasive ventilation (NIV) could mean a little more “own space,” more possibilities, and a life with more freedom, as explained in the following quote: “I would like if she could get rid of the tracheotomy, then you can travel abroad and so. Because it is the tracheotomy that requires all the PCAs.” However, the reverse was also true—the sibling’s illness became the reason why the family had the opportunity to go on a holiday for the first time: “My sister had ‘Your Big Day,’ so we will go abroad for the first time.” The physical limitations of the ill sibling also required compensating for the sibling’s limited physical ability to do different activities: “I usually help him with things, actually a little bit of everything. There are those little things that you do. If we had not helped him, he would not have been able to do anything.” The physical dimension was experienced as a protective assignment, a kind of mission to make sure that nothing happens to the HMV-assisted sibling. This mission became natural when being siblings: “So, I’m always trying, of course, to make sure nothing happens to him, you’ll do that automatically as brothers.”

Having an HMV-assisted sibling meant being forced to grow up fast and, perhaps, to be more mature than one’s peers:
My sister takes quite a lot of time, so I do not get as much time. That is why I take care of myself, which is why I have learned that. My parents cannot always help me. Then, I take care of myself.

It was about being shaped into a human being, (i.e., a psychical dimension) who has openness and acceptance to a person or to a life that might be a little different, as explained in the following quote:

In school, for example, I can talk to people who may look a little different than everyone else. They may have some disease. I think I dare to talk to them and so on. I may be able to relate to them because I have someone that is a little different in my vicinity. Having a sibling that is different makes you a little different.

Having an HMV-assisted sibling developed into something that one became used to (but still not) in relation to having a lot of experiences and different situations. Being used to in this sense sometimes meant getting used to a sibling’s convulsions at home in the middle of the night, but it could just as well mean never getting used to when there was a problem with your sibling’s tracheotomy at nights and, hence, always waking up and standing near them. It involved getting used to medical devices at home and having PCAs within the family for as long you could remember. But it also meant to never really be used to living in a crowded home that did not allow friends to be invited or had PCAs around at night.

Comprehensive Understanding
As a final step of the analysis, a comprehensive understanding of the whole text was formulated. The interpreted whole showed that the meaning of having a sibling with HMV was understood as having a complex and profound intertwined bond that links to the past, the present, and future. This different but not strange bond shaped you to become a person prepared to fight for diversity and defend your sibling’s right to be themselves. Much like a brave and loyal knight with armor and a powerful, but vulnerable, empire on the inside. This profoundly intertwined bond shaped character traits such as maturity, empathy, and knowledge. Nonetheless, it has additionally created worries (about periods of disease exacerbation), concerns (about parents’ concerns), responsibility (for the ill sibling), being forced to grow up fast, and having limited time and space with one’s parents. This way of being can be compared with Gadamer’s (1996) way of characterizing health as a kind of equilibrium condition, a rhythmic experience when life is in harmony, and people find their own paths in consonance with the world.

Discussion
This current study revealed new important knowledge, which adds to existing knowledge, about siblings’ everyday life experiences when having an HMV-assisted brother or sister. The results highlight that these siblings live in a unique family circle where they have become used to having limited time and space with parents and feelings of worries and concerns for their sibling and parents. Despite this, they expressed that this experience had equipped them with an ability to listen to the needs of others with a great deal of acceptance, understanding, and knowledge. We noticed that temporality was present in the participants’ testimonies, which we describe as the past, the present, and the future. Phenomenological hermeneutics aims at a description and interpretation of a possible existence and belonging in the world (Ricoeur, 1998). Ricoeur (1988, 1998) used the concept of historicity and claimed that being human is being one’s own history and narrative. In the comprehensive understanding, we have tried to bring the interpreted history of being a sibling to an HMV-assisted child into words.

The participants’ narratives were much about a daily life where there is a sibling with a major disability who also requires respiratory support. The ventilator treatment itself was not considered a great challenge, which is similar to previous research findings (Gonzalez et al., 2017; Graham, Rodday, Weidner, & Parsons, 2016; Pelentsov, Fielder, & Esterman, 2016). However, having a sibling with tracheotomy seemed to be extra challenging because, at all times of the day, this situation required attentiveness from parents and PCAs in the family’s home. This situation placed additional demands on parents, which in turn affects the sibling’s everyday life, which previous research with this population has also shown (Amin et al., 2017; Wilfond, 2014).

Woodgate and colleagues (2016) noted that, when interviewing the siblings of children with complex care needs, despite the participants’ strong sense of love for their siblings, they found it difficult when their family avoided activities to maintain safety and comfort.

Overall, in our study, even if the siblings’ lives were not always as they wanted them to be, the siblings had become accustomed to their situation, making it into a kind of habit. They appeared to be very loyal to their siblings and understanding of their parents’ situation, explaining that they had learned to handle many of their own issues alone because their parents often were occupied. This is in line with Carnevale and colleagues (2006) findings, where siblings expressed that their situation did not always suit them, but they understood what their parents were experiencing. Stoneman (2005) found that siblings in well-functioning families interpret differential parenting as merely a required response to the special needs of their sibling rather
Methodological Reflections

According to Ricoeur (1976), there is more than one way of constructing and interpreting a text. We do not claim that we have found one single truth. Rather, we present what we consider is one of the best interpretations that support our data about a possible belonging in the world when being the sibling of an HMV-assisted child. The structural analysis helped us view the text as objectively as possible (Lindseth & Norberg, 2004) and validate our interpretations. Quotations are presented to illustrate subthemes and themes and strengthen trustworthiness in the results. The authors represent an interdisciplinary research team consisting of a physician specialized in HMV care for children, two intensive care nurses, one pediatric nurse, and one speech and language therapist specialized in qualitative methods. We reached a negotiated consensus through discussions during the analysis process. Moreover, three of the authors are experienced researchers in the HMV area. To strengthen credibility, an ongoing discussion about understanding occurred. The 10 interviews were performed by first author (Å.I.-S.). All five authors have read and interpreted the interview text and discussed the themes and subthemes. Research seminars, discussions, and reflections facilitated a critical stance to ensure rigor (Morse et al., 2002). Regarding transferability, the findings may be valuable when it comes to other groups of siblings who have brothers or sisters with long-term illnesses. All participants’ families probably had a well-functioning relationship with the health care system, which is where the participants were gathered. This fact could have affected the goal of achieving a representative sample and we may have missed families that had a complicated relation to health care. This can be regarded as a limitation. However, the siblings spoke both about positive and negative experiences. Only those who spoke and understood the Swedish language were asked about participation, which must be regarded as a limitation. However, the other groups of siblings who have brothers or sisters with long-term illnesses are difficult to reach within many areas, and it is important to strive for solutions to meet this challenge.

Implications for Practice With Families

The results confirm that it is important for siblings to have the opportunity to be close with their family if their HMV-assisted sibling is admitted to the hospital for a long period of time. It is of great significance for health care providers to offer siblings tailored support, for example, for younger siblings the opportunity to play, and for older siblings the opportunity to create their own private zone where they can be themselves when staying at hospital residences. Health care providers often establish a close, long-term relationship with these families and can establish a sensible and open-hearted relationship with the siblings, being responsive if they are in need of a professional interlocutor. When the sibling is living with his or her family at the hospital, there
might be fun and stimulating activities for the sibling to attend. But when the sibling and family are home again and are going through everyday life, it can be challenging to get attention and time from the parents. The siblings also reported a need to have a break from the HMV-assisted child and spending time with parents, with no PCAs in the house. Therefore, the parents should be acknowledged and supported to meet the sibling’s wishes of being seen on their premises. It may be helpful for health care providers to adopt the competencies suggested by the International Family Nursing Association (2015) Position Statement on Generalist Competencies for Family Nursing Practice that include a focus on family strengths to support siblings’ individual growth, management of health, and transitions in life.

Conclusion

The current study contributes new knowledge that comes from an insider’s perspective about the meaning of having an HMV-assisted sibling. The research uncovered that siblings have a complex and profound intertwined sibling bond that links to the past, present, and future. How can health care professionals help siblings strengthen this profound intertwined bond so that it does not become too tense and overwrought? The findings of this study provide valuable information for family-focused care of HMV-assisted children and their families where well siblings must be given attention and support. Good quality care can support siblings in developing their own unique strengths and self-growth. Further research is needed about what happens to adult siblings when this profound intertwined bond progresses.

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ABSTRACT

**Aim:** The population of children requiring home mechanical ventilation (HMV) has grown in number and complexity in the past few decades. This study explored the health related quality of life (HRQoL), family functioning and sleep of parents to children with HMV in Sweden. A secondary aim was to explore the impact on HRQoL, family functioning and sleep of selected potential determinants.

**Methods:** Questionnaires were completed by 85 parents to 55 children receiving HMV in Sweden. Parents were identified via the Swedish national quality register for oxygen and home respiratory treatment and were invited to participate by health care professionals between December 2016 and December 2018.

**Results:** Parents to children with HMV reported lower HRQoL and family functioning in comparison to previous research. One of four parents reported moderate or severe clinical insomnia. Parents HRQoL and family functioning were predicted by the child’s HMV mode and sleep. There were no differences between mothers and fathers for overall HRQoL, family functioning or sleep reports.

**Conclusion:** In comparison with previous research this study showed that parents to children with HMV reported low HRQoL and family functioning. Having a child with tracheotomy and having sleep problems were related and predicted parents reported HRQoL and family functioning.

**Keynotes**

- The population of children with home mechanical ventilation (HMV) have grown in number and complexity.
- Parents to children receiving HMV reported lower health related quality of life (HRQoL) and family functioning in comparison to previous research addressing parents to children with long-term conditions.
One of four parents reported insomnia. Sleep quality and the child’s HMV mode predicted HRQoL. No differences between mothers’ and fathers’ overall reports were found.

Keywords
Family functioning, health related quality of life, home mechanical ventilation, parents, sleep

INTRODUCTION

In the past few decades, more children have received home mechanical ventilation (HMV) (1); today approximately 300 children in Sweden are receiving HMV (2). These children constitute a diverse group, regarding not only their diagnosis and breathing problems but also their complex healthcare needs (3), which results in medical fragility and functional limitations. HMV can be delivered invasively via a tracheostomy tube or noninvasively (NIV) and for only part of the day, typically during sleep, or continuously for 24 hours a day (4). Although HMV supports respiratory function, it does not alleviate other conditions; thus, a wide variety of unique and complex care services are delivered in the homecare environment, outside of a hospital setting (1).

A consequence of the homecare setting is the parents’ extended medical responsibility. Health related quality of life (HRQoL) and family functioning are affected among parents (5, 6) to children with HMV treatment. Families have in many ways taken the responsibility for required advanced homecare, which often involves high vigilance and administering skilled care both day and night (7). Parents to children with congenital central hypoventilation syndrome frequently experience a disturbed night sleep (8), and several aspects of a perceived HRQoL are affected in parents to children receiving HMV (9). Mothers, in particular, have reported poor sleep quality (10). Parents often go to bed in a state of readiness, prepared to respond to alarms from medical devices (9). Some parents wake up early in the morning to perform technical care and routines (7). Emotional and cognitive symptoms of stress related to poor sleep can pose a threat to a person’s wellbeing (11), their relationships and their capacity to maintain vigilance and good quality of care (12).

Caring for a child receiving HMV often changes and effects the parents’ role and functioning within the family (13) as well as their ability to perform daily activities, handle family finances (1) and care for the wellbeing of other family members (14). Research highlights that it is often the mother who has the main responsibility for the child’s care (15) and, frequently, when it becomes impossible for both parents to keep up their employment it is often the mother who gives up her professional life and career (13). Swedish legislation supports parents to work as a personal care assistant (PCA) for their own child (16). If and how this affects parents ability and wishing to work outside home is unknown in this context.

The complex needs of a child with HMV affects the parents’ HRQoL, functioning (13) and sleep (9). The parental role often changes to include nursing dimensions (13) and parents who are burdened and stressed may experience significant negative physical and emotional consequences to their own health (17). Focusing on parental HRQoL, family functioning and sleep from a parental and a family system perspective is vital when there is an inextricable link between parental HRQoL and the child’s wellbeing (18). Several aspects, for instance HMV mode and unique care situations have not been explored in relation to parents’ sleep and impact on HRQoL and family functioning (7). To the best of our knowledge, this study is the first that explores several aspects that may affect parents’ HRQoL, family functioning and sleep in the Swedish context. The aim of the study was therefore to explore HRQoL, family functioning and sleep in parents to children
Parents and home mechanical ventilation receiving HMV in Sweden. A secondary aim was to explore the impact on HRQoL, family functioning and sleep of selected potential determinants.

PATIENTS AND METHODS

Design
A cross-sectional study was designed using parent reported data for exploring parents HRQoL, family functioning and sleep in relation to being a mother or father. The child’s HMV mode, defined as 1) tracheotomy, 2) non-invasive ventilation (NIV) or 3) continuous positive airway pressure (CPAP), and if the parents were employed as a PCA or in paid work outside home was also related to the Peds QL Family Impact Module and Insomnia Severity Index (ISI) reports.

Participants and settings
In total 88 participants were included; 85 participants from 55 families responded to the questionnaires. Paired data, from both mothers and fathers, were obtained from 30 families; altogether 60 parents living together with each other. Inclusion criteria comprised being a parent to a child of 0-18 years, with HMV therapy. The exclusion criterion was an inability to speak or understand Swedish. The parents, who fulfilled the study inclusion criteria, were consecutively invited to participate by healthcare professionals between December 2016 and December 2018. Children with HMV in Sweden (n=300) are usually treated in a respiratory clinic, and these clinics were identified via a Swedish National Quality Register for Oxygen and Home Respiratory Treatment, Swedevox (2). Actual clinics were informed about the study and were requested to ask parents about their participation. The study was approved by the Regional Ethical Review Board of Gothenburg, Sweden (Ref. No. 354-15). Parents were informed about the aim of the study, confidentiality and the voluntariness to participate which followed the guidelines of the Declaration of Helsinki (19).

Measures
Peds QL Family Impact Module was developed to assess what impact a child’s chronic health condition has on the parents (20). The questionnaire consists of 36 items and three summary scores can be derived: the Total Score (36 items), the Parent HRQoL Summary Score (20 items) and the Family Functioning Summary Score (8 items). The Parent HRQoL Summary Score is calculated by averaging the items from four scales measuring physical functioning (6 items), emotional functioning (5 items), social functioning (4 items) and cognitive functioning (5 items). The Family Functioning Summary Score is a summative average of the following scales: daily activities (3 items) and family relationships (5 items). The Total Score is calculated by averaging the above 28 items together with items from the communication scale (3 items) and worry scale (5 items). The response format is a five-point Likert scale, in which items are scored from never a problem (0) to always a problem (4). The scores are reversed and transformed into a zero to 100 scale, in which higher scores indicate better functioning. The validity and reliability are well documented (21). The Family Impact Module has been translated into Swedish and used in Swedish research (22). A linguistic validation into Swedish was performed on 109 participants. The subscales were assessed for internal consistency reliability using Cronbach’s alpha. The Total Score, the Parent HRQoL Summary Score and the Family Functioning Summary Score achieved values greater than 0.70 (23).

ISI is a seven-item self-reported questionnaire with documented reliability and validity for detecting insomnia (24). ISI has been translated into Swedish and used in population studies both nationally (25) and internationally (26). The ISI evaluates the severity of sleep-onset (initial), sleep maintenance (middle) and early morning waking problems (terminal) as well as sleep satisfaction, interference with daily functioning, noticeability of sleep problems and distress caused by sleep problems the last two weeks. The items are ranked on a five-digit Likert scale in which the
Parents and home mechanical ventilation

items are scored from never a problem to always a problem and merged into a total score, which range from zero to 28; a score of ≥15 indicate clinically significant insomnia. The questionnaires were completed individually by each parent during a hospital visit and were placed in a sealed box.

Statistical analyses

Descriptive statistics were used for background characteristics, Peds QL and ISI. Data were tested for normality, and since the criteria were reached, parametric methods were used for analyses. The Paired t-test for comparison of mean values from Peds QL; Total score, Parent HRQoL Summary Score, Family Functioning Summary Score and ISI score were used to analyse differences between the cohabiting parents (n=60). Differences between parents’ HRQoL Summary Score and Family Functioning Summary Score were explored in relation to HMV mode using one-way ANOVA. ISI reports were dichotomized, <15 and ≥15, according to the defined cut-off values (27). The association between the mother’s and father’s reports were analysed by chi-square test in relation to HMV mode, working as PCA or working as an employee outside the home. Linear multiple regressions with backward selection were used to predict the relationships between the dependent variables parents’ HRQoL Summary Score and Family Functioning Summary Score. The independent variables were gender (10), parents working as PCA or not (6), parents having paid work outside the home or not (6), HMV mode (11) and ISI total score (14). The criteria for linear multiple regression (normality, linearity and homoscedasticity) (28) were controlled and satisfactorily met. Regression models were evaluated using the adjusted R². A significance level was set to p < .05. IBM SPSS™ Statistics version 25 was used for the analyses.

RESULTS

The background characteristics of parents and children are presented in Table 1 and 2. A total of 88 parents to 55 children were recruited to respond to the questionnaires. Paired data, from both mothers and fathers, were obtained from 30 families, altogether 60 parents living together with each other. Three questionnaires were incomplete and were excluded which left 85 for the analysis: 45 from mothers and 40 from fathers. Both parents filled in the questionnaire for 30 children, in the other cases (n=25), either the mother or father did. The parents’ mean age was 41 years; for the mother, it was 41 years, and for the fathers, it was 43 years. The children’s mean age was 8.5 years, and the mean time with HMV treatment was six years and eight months. The largest group (n=31) had NIV treatment.

Table 1 Characteristics of parents to children receiving HMV

<table>
<thead>
<tr>
<th>Participants (n=85)</th>
<th>n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mothers</td>
<td>45(53)</td>
</tr>
<tr>
<td>Fathers</td>
<td>40(47)</td>
</tr>
<tr>
<td><strong>Age (n=85)</strong> Mean (SD)</td>
<td></td>
</tr>
<tr>
<td>Parents</td>
<td>41(6)</td>
</tr>
<tr>
<td>Mothers</td>
<td>41(6)</td>
</tr>
<tr>
<td>Fathers</td>
<td>43(6)</td>
</tr>
<tr>
<td><strong>Education (n=85)</strong> n (%)</td>
<td></td>
</tr>
<tr>
<td>Elementary school father/mother</td>
<td>2 (5) / 3 (6)</td>
</tr>
<tr>
<td>High school father/mother</td>
<td>18 (46) / 20 (44)</td>
</tr>
<tr>
<td>University father/mother</td>
<td>19 (48) / 20 (44)</td>
</tr>
<tr>
<td>Other father/mother</td>
<td>0 (0) / 2 (4)</td>
</tr>
<tr>
<td><strong>Employment (n=85)</strong> n (%)</td>
<td></td>
</tr>
<tr>
<td>Employment father (n=38) / mother (n=44)</td>
<td>35 (95) / 36 (81)</td>
</tr>
<tr>
<td>Fulltime work father (n=38) / mother (n=44)</td>
<td>26 (68) / 20 (22)</td>
</tr>
<tr>
<td>Father or mother working as a PCA</td>
<td>49 (60)</td>
</tr>
<tr>
<td>Father working as PCA father (n=38) / mother working as a PCA (n=44)</td>
<td>21 (55) / 28 (63)</td>
</tr>
<tr>
<td><strong>Family characteristics (n=55)</strong> n (%)</td>
<td></td>
</tr>
<tr>
<td>Parents living together with each other</td>
<td>80 (71)</td>
</tr>
<tr>
<td>Having two or more children (n=53)</td>
<td>44 (83)</td>
</tr>
<tr>
<td>Having a PCA employed (n=53)</td>
<td>46 (86)</td>
</tr>
<tr>
<td>Having two PCAs in tandem (n=53)</td>
<td>14 (27)</td>
</tr>
<tr>
<td>PCA hours / week (n=55) mean (SD)</td>
<td>123 (72)</td>
</tr>
<tr>
<td>PCA = Personal care assistant</td>
<td></td>
</tr>
</tbody>
</table>
The impact on parents’ HRQoL and family functioning is presented in Table 3. The mean values within the HRQoL dimensions ranged between 49 and 70; the lowest values were found for the mothers’ physical, emotional and social functioning, and the highest values were for fathers’ cognitive functioning. Overall, and for most dimensions, there were no significant differences within the couples’ HRQoL. The only differences found were within the physical and cognitive functioning (Table 3).

The impact on family functioning dimensions ranged between 40 and 57; the lowest values were found for the mothers’ daily activities, and the highest values were found for family relationship rated by the fathers (Table 3). There were no difference within the couples’ ratings of family functioning.

The parents’ reports of insomnia are presented in Table 4. Dichotomization of the scale scores showed that 25% of parents reported ISI ≥15, indicating moderate to severe insomnia (Table 4). The chi-square test showed no differences between mothers and fathers perceived sleep problems.

### Table 2 Characteristics of children receiving HMV

<table>
<thead>
<tr>
<th>Characteristics of children receiving HMV (n=55)</th>
<th>Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>8.2 (4)</td>
</tr>
<tr>
<td>Diagnostic category (n=52)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Neuromuscular disease</td>
<td>12 (23)</td>
</tr>
<tr>
<td>Central apnoea</td>
<td>11 (21)</td>
</tr>
<tr>
<td>Sleep apnoea</td>
<td>5 (9)</td>
</tr>
<tr>
<td>Chromosomal aberrations</td>
<td>9 (17)</td>
</tr>
<tr>
<td>Other</td>
<td>15 (28)</td>
</tr>
<tr>
<td>HMV mode (n=53)</td>
<td>n (%)</td>
</tr>
<tr>
<td>CPAP</td>
<td>13 (24)</td>
</tr>
<tr>
<td>NIV</td>
<td>31 (58)</td>
</tr>
<tr>
<td>Tracheotomy</td>
<td>9 (17)</td>
</tr>
<tr>
<td>Time with HMV (n=52)</td>
<td>n (%)</td>
</tr>
<tr>
<td>HMV part of the day</td>
<td>13 (25)</td>
</tr>
<tr>
<td>HMV day or night</td>
<td>5 (9)</td>
</tr>
<tr>
<td>HMV day and night</td>
<td>28 (53)</td>
</tr>
<tr>
<td>HMV 24 hours</td>
<td>8 (15)</td>
</tr>
<tr>
<td>Number of months with HMV treatment (n=53)</td>
<td>mean (SD)</td>
</tr>
<tr>
<td>.73 (54)</td>
<td></td>
</tr>
<tr>
<td>Level of education (n=52)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Pre-school</td>
<td>8 (16)</td>
</tr>
<tr>
<td>Elementary school</td>
<td>31 (60)</td>
</tr>
<tr>
<td>High school</td>
<td>5 (10)</td>
</tr>
<tr>
<td>Home teaching</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>

*CPAP = Continuous positive airway pressure; NIV = Noninvasive ventilation

### Table 3 Mean values between reported health related-quality of life and family functioning from the total sample and differences between mothers and fathers living together with each other. (Parents Peds QL Family Impact Module Version 2.0)

<table>
<thead>
<tr>
<th>Scales</th>
<th>Items</th>
<th>Parents (total)</th>
<th>Mothers</th>
<th>Fathers</th>
<th>Differences between parents*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (SD)</td>
<td>n</td>
<td>Mean (SD)</td>
<td>n</td>
<td>Mean (SD)</td>
</tr>
<tr>
<td>Total score</td>
<td>26</td>
<td>64</td>
<td>54(15)</td>
<td>45</td>
<td>51(20)</td>
</tr>
<tr>
<td>Parent health related quality of life summary score</td>
<td>20</td>
<td>64</td>
<td>52(15)</td>
<td>45</td>
<td>47(20)</td>
</tr>
<tr>
<td>Physical functioning</td>
<td>6</td>
<td>64</td>
<td>54(22)</td>
<td>45</td>
<td>49(22)</td>
</tr>
<tr>
<td>Emotional functioning</td>
<td>5</td>
<td>64</td>
<td>51(21)</td>
<td>45</td>
<td>49(21)</td>
</tr>
<tr>
<td>Social functioning</td>
<td>4</td>
<td>64</td>
<td>51(25)</td>
<td>45</td>
<td>49(27)</td>
</tr>
<tr>
<td>Cognitive functioning</td>
<td>5</td>
<td>64</td>
<td>62(26)</td>
<td>45</td>
<td>54(29)</td>
</tr>
<tr>
<td>Communication</td>
<td>3</td>
<td>64</td>
<td>56(24)</td>
<td>45</td>
<td>55(23)</td>
</tr>
<tr>
<td>Worry</td>
<td>5</td>
<td>65</td>
<td>52(20)</td>
<td>45</td>
<td>53(20)</td>
</tr>
<tr>
<td>Family functioning summary score</td>
<td>8</td>
<td>65</td>
<td>51(23)</td>
<td>45</td>
<td>50(23)</td>
</tr>
<tr>
<td>Daily activities</td>
<td>3</td>
<td>65</td>
<td>43(26)</td>
<td>45</td>
<td>40(26)</td>
</tr>
<tr>
<td>Family relationships</td>
<td>3</td>
<td>65</td>
<td>45(26)</td>
<td>45</td>
<td>45(25)</td>
</tr>
</tbody>
</table>

*Summarising all items in the questionnaire + Summarising functional problems in physical, emotional, social and cognitive scale + Summarising daily activities and family relationships + Differences between mothers and fathers (n=60) living together with each other (n=30).

### Table 4 Reported level of insomnia in parents to children receiving HMV. Differences are presented between parents living together with each other (n=30) (Insomnia Severity Index).

<table>
<thead>
<tr>
<th>Scale</th>
<th>Score</th>
<th>Parents</th>
<th>Mothers</th>
<th>Fathers</th>
<th>Differences between parents*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>n=82</td>
<td>n=42</td>
<td>n=40</td>
<td>n=40</td>
</tr>
<tr>
<td>n(%)</td>
<td>n(%)</td>
<td>n(%)</td>
<td>n(%)</td>
<td>p-value</td>
<td></td>
</tr>
<tr>
<td>Absence of insomnia</td>
<td>0-7</td>
<td>29(35)</td>
<td>12(28)</td>
<td>17(42)</td>
<td></td>
</tr>
<tr>
<td>Subthreshold insomnia</td>
<td>8-14</td>
<td>32(39)</td>
<td>18(42)</td>
<td>14(35)</td>
<td></td>
</tr>
<tr>
<td>Moderate insomnia</td>
<td>15-21</td>
<td>17(20)</td>
<td>10(23)</td>
<td>7(17)</td>
<td></td>
</tr>
<tr>
<td>Severe insomnia</td>
<td>22-28</td>
<td>4(4)</td>
<td>2(4)</td>
<td>2(5)</td>
<td></td>
</tr>
<tr>
<td>ISI total score ≤15</td>
<td>&lt;15</td>
<td>6(17)</td>
<td>3(7)</td>
<td>3(7)</td>
<td></td>
</tr>
<tr>
<td>ISI total score &gt;15</td>
<td>≥15</td>
<td>21(28)</td>
<td>12(28)</td>
<td>9(23)</td>
<td></td>
</tr>
</tbody>
</table>

*p-value 0.227 Differences between mothers and fathers, living together with each other (n=30)
Parents and home mechanical ventilation

There were no associations between parents’ ISI reports, the child’s HMV mode (X[I3, N=82] = 1,270, p=0.788) and whether the parents worked as a PCA (X[I2, N=82] = 1,450 p=0.518) or had a paid work outside the home or not (X[I2, N=82] = 0.962, p=0.673). There was a difference between the parents’ HRQoL Summary Score (F [2, 81] = 4.641 p=0.012) and the Family Functioning Summary Score (F [2, 82] = 3.737 p=0.028) in relation to HMV mode. A Tukey post-hoc test revealed significant differences between the HMV modes tracheotomy, NIV and CPAP according to the parents’ HRQoL Summary Score and Family Functioning Summary Score. Being parent to a child with tracheotomy had more impact on HRQoL (m=38.9) and Family Functioning (m=37.5) compared to NIV and CPAP according to the parents’ HRQoL Summary Score and Family Functioning Summary Score. Being parent to a child with tracheotomy had more impact on HRQoL (m=38.9) and Family Functioning (m=37.5) compared to NIV (HRQoL Summary Score [m=52.9], Family Functioning Summary Score [m=54.3]) and CPAP (HRQoL Summary Score [m=58.8] and Family Functioning Summary Score [m=57.6]).

The multiple linear regression analyses (Table 5 and 6) showed that the basic model with the five independent variables was a significant predictor of parents’ HRQoL Summary Score (R=0.682), explaining 46% of the variance. The adjusted R² value in the regression model indicated that 45% of the variability in the dependent variable Parent HRQoL Summary Score was predicted by the child’s HMV mode and ISI reports (Table 5). The child’s HMV mode and ISI total score were also found to predict the Family Functioning Summary Score, explaining 21% of the variance (Table 6).

### Table 5: Multiple regression models with Parent HRQoL Summary Score (n = 85) as the dependent variable

<table>
<thead>
<tr>
<th>Predictor</th>
<th>B</th>
<th>Std. Error</th>
<th>t</th>
<th>Sig.</th>
<th>Adjusted R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>Model A</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender, mother or father</td>
<td>-3.583</td>
<td>0.196</td>
<td>18.562</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Parent working as a PCA or not</td>
<td>-2.303</td>
<td>0.132</td>
<td>18.736</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Parent has paid work outside the home or not</td>
<td>-2.617</td>
<td>0.134</td>
<td>17.317</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>HMV mode</td>
<td>-1.519</td>
<td>0.131</td>
<td>11.396</td>
<td>&lt;0.001</td>
<td></td>
</tr>
<tr>
<td>Total score</td>
<td>-1.400</td>
<td>0.130</td>
<td>10.502</td>
<td>&lt;0.001</td>
<td></td>
</tr>
<tr>
<td>Adjusted R²</td>
<td>0.451</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

### Table 6: Multiple regression models with Family Functioning Summary Score (n = 85) as the dependent variable

<table>
<thead>
<tr>
<th>Predictor</th>
<th>B</th>
<th>Std. Error</th>
<th>t</th>
<th>Sig.</th>
<th>Adjusted R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>Model A</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender, mother or father</td>
<td>-3.583</td>
<td>0.196</td>
<td>18.562</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Parent working as a PCA or not</td>
<td>-2.303</td>
<td>0.132</td>
<td>18.736</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Parent has paid work outside the home or not</td>
<td>-2.617</td>
<td>0.134</td>
<td>17.317</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>HMV mode</td>
<td>-1.519</td>
<td>0.131</td>
<td>11.396</td>
<td>&lt;0.001</td>
<td></td>
</tr>
<tr>
<td>Total score</td>
<td>-1.400</td>
<td>0.130</td>
<td>10.502</td>
<td>&lt;0.001</td>
<td></td>
</tr>
<tr>
<td>Adjusted R²</td>
<td>0.451</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

PCA = Personal care assistant; HMV = Home mechanical ventilation; HMV mode = Continuous positive airway pressure (CPAP), Noninvasive ventilation (NIV) and Tracheotomy; ISI = Insomnia severity index
DISCUSSION

This study showed that parents to children with HMV in Sweden reported low HRQoL. In another Swedish study (22), in which Peds QL Family Impact module was used to investigate HRQoL in parents to children with Type 1 Diabetes, the mothers and fathers reported better functioning within the HRQoL and family functioning area. Reports on HRQoL and family functioning in our study are comparable to an American study (29) in which parents to children dependent on medical technology responded to Peds QL Family Impact Module. The parents’ reports in our study might be understood from the medical status of their children, which often requires advanced homecare that parents in many ways are responsible for. Previous research has showed that not only the severity of the child’s underlying medical condition but also how the parents cope with the situation plays an important role for HRQoL (5, 6, 11). Parents in the HMV context with good social support had better mental health and cognitive, social, and family functioning (30).

Additionally, our results showed that the child’s HMV mode and the parents’ sleep quality predicted parents’ HRQoL. The size and direction of this relationship underscored that being a parent to a child with tracheotomy and having sleep problems significantly affected HRQoL. This seems understandable, since children with tracheostomy require continuous monitoring day and night and may also have complex disease conditions that involve multiple technologies (31). Previous research has also strengthen such a relationship, and has showed that having a child being dependent on medical technology increased the parental care burden, resulting in daily fatigue and little energy for household tasks or social activities (29). Sleep problems had the strongest relationship with HRQoL, which may not be surprising, since previous research has described parental caregiving in this context as comparable to working a rotating shift, different nights of the week. This has been shown to negatively affect health outcomes (9), which deserves attention, considering that 25% of the parents in this study reported moderate or severe clinical insomnia.

Mothers’ reported clinical insomnia to a greater extent than the fathers. This trend may indicate that having a medically fragile child with functional limitations may affect mothers’ HRQoL to a greater extent. However, no significant differences between mothers’ and fathers’ HRQoL, family functioning or sleep were found. Statistical power might be an explanation, but the results may also reflect that parents have to support each other in order to make ends meet. Consequently, the situation may be demanding and affect HRQoL, family function and sleep for both parents, regardless of gender.

The study’s results can be understood from the Swedish context, where the legislation (16) make it possible for families with a child receiving HMV to have PCAs. Parent reaction patterns can hardly be generalized without considering national differences in health care provision and variations in the possibilities for PCAs. Differences in health care system exist and may affect the possibility to generalise results. In this study PCAs were present in almost all of the families, and more than half of the fathers and mothers were working as a PCA themselves. Almost all fathers and mothers also had an occupational activity outside of home. However, whereas 68% of the fathers worked full time, only 22% of the mothers did (Table 1). The latter difference is in line with Swedish society in general; mothers work part time more often than fathers do when they become parents (32).

One of this study’s strengths is that the numbers of mothers and fathers was similar, which is in contrast to other studies within this context. The number of parent participants in total is considered fair (n=85) since a total of approximately 300 children receive HMV in Sweden. Parents from all parts of Sweden were also
Parents and home mechanical ventilation

included in the study. This mirrors regional variations in type of residence, healthcare organisations, accessibility to hospital and equipment support. The results have the potential to be generalised to other groups of children with HMV treatment and various long-term illnesses, since parents to children with a wide spectrum of diagnoses have responded to the questionnaire. This study also has some limitations. There were no matched healthy control group, which further could have elucidated whether parents to children with HMV are impacted regarding HRQoL and family functioning. Yet the PedsQL Family Impact Module was developed to assess what impact a child’s chronic health condition has on the parents, and from our point of view, this limits its use to clinical samples. There are scores from healthy samples reported in previous studies, but these studies aim to explore the psychometric properties of the instrument and not to develop reference data (21, 33). Though, it had been helpful to have cut off scores for this measure when clarifying the effect that a child’s chronic disease has on parents. This paves the way to further research. Future research could also identify variables not evaluated in this study, which may be predictors affecting parent reports. For example being single parent, education and income, housing situation and the child’s disease severity and age.

The participating parents were to a large extent well educated and Swedish speaking; they likely had a well-functioning relationship with the healthcare system. These factors could have contributed to a bias regarding sociodemographic conditions in this context.

CONCLUSION

In comparison with previous research, this study showed that parents to children with HMV in Sweden reported low HRQoL and family functioning. One of four parents reported moderate or severe insomnia. The child’s HMV mode and parents’ sleep quality were related and predicted the parents’ HRQoL. No significance difference between the mothers’ and fathers’ reports was found. Findings in this study indicate that a multicompetent and responsive support addressing both mothers and fathers HRQoL, family functioning and sleep habits seems to be important.

Abbreviations

CPAP, Continuous positive airway pressure; HRQoL, Health related quality of life; ISI, Insomnia severity index; NIV, Noninvasive ventilation; PCA, Personal care assistant; PedsQL, Paediatric Quality of Life

CONFLICTS OF INTEREST

The authors have no conflict of interest to declare.

FUNDING

No external funding was received for this study.

REFERENCES


