Home mechanical ventilation in complex chronic disease in children – what is the impact on the sleep quality of the caregivers?

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Raquel Santos Gonçalves raquel.santos.goncalves@gmail.com
Hospital Pediátrico de Coimbra, Centro Hospitalar e Universitário de Coimbra
Corresponding Author
ORCiD: 0000-0003-4090-4808

Cristiana Martins
Universidade de Coimbra Faculdade de Medicina

Ana Borges
Centro Hospitalar e Universitario de Coimbra EPE

Núria Madureira
Centro Hospitalar e Universitario de Coimbra EPE

Cândida Cancelinha
Centro Hospitalar e Universitario de Coimbra EPE

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Abstract

Background

The role caregivers of children with complex chronic disease play may be linked to important sleep disturbances, which may threaten the long-term quality care to their children.

Aim

Evaluation of the sleep quality of the caregivers of children with complex chronic disease under home mechanical ventilation.

Design

Observational and analytic study (October 2018 – January 2019).

Setting/Participants

We assessed 33 caregivers of patients with complex chronic disease under home mechanical ventilation for ≥3 months, followed in the Pneumology Clinic of a tertiary paediatric hospital. Evaluation instrument: Pittsburgh Sleep Quality Index (Portuguese version).

Results

Most of the caregivers were mothers (88%), median age of 40 years. Most of the children were male (70%), median age of 11 years. The main diagnostic group was genetic/polymorphic syndrome (46%); 52% belonged to group 2 (Association for Children with Life-Threatening or Terminal Conditions). There was a discrepancy between the subjective sleep quality ("Good" in 64%) and the Pittsburgh Sleep Quality Index score ("Poor" in 72%), with significant percentage of daytime sleepiness/dysfunction (75%). The increase in daily hours of ventilation seems to lead to a lower sleep quality (p = 0.669). Caregivers with sleep disorder had lower sleep quality values.

Conclusions
The sleep quality of the caregivers of children under home mechanical ventilation has similar values to specific groups described in literature, but lower than that of the healthy Portuguese population. It is essential to raise the professionals’ awareness for the evaluation of sleep quality of caregivers, since it may not be adequately perceived by them.

Background

In the last few decades, there has been a substantial rise in the number of children and adolescents depending on home mechanical ventilation, due to an increase in the number of patients surviving critical illness and due to technological advances.(1–4) Home mechanical ventilation allows a prolongation of life, palliation of symptoms and an improvement in the quality of life.(1) With this, the care of many of these children has shifted from professionals in the hospital to parents in the family home, and society increasingly relies on the caregivers of dependent children to provide highly skilled and vigilant care in their homes 24 hours a day.(2,3)

The caregivers of children who depend on medical technology at home have been found to experience numerous negative outcomes, including high levels of physical, emotional, financial and family stress.(5) The extraordinary responsibility that caregivers undertake may be linked to chronic sleep disturbance, which may threaten their ability to provide long-term quality care for their children.(2)

Sleep is recognized as a complex biobehavioral process described as “a reversible behavioral state of perceptual disengagement from, and unresponsiveness to, the environment”.(6) Sleep, in adequate quantity and good quality, is considered necessary for an overall health and optimal daytime performance. Shorter sleep duration and higher nocturnal awakenings have been reported, when comparing sleep in caregivers of children with physical disabilities to those of typically developing children.(2) Chronic sleep
deprivation is considered a major public health concern and has been associated with
caregiver stress and depression.(2,7)

In 1993, a home mechanical ventilation program for children and adolescents with
chronic respiratory failure began in our hospital. It has already supported hundreds of
patients with a positive impact in patients and caregivers’ lives.(4)

Methods

Observational and analytic study performed between October 2018 and January 2019. The
study group was defined based on a convenience sample consisting of parents/caregivers
of children/adolescents (0–17 years) with complex chronic disease, followed in the
Pneumology Clinic of our hospital. The inclusion criteria were parents/caregivers of
children/adolescents with complex chronic disease under home mechanical ventilation for
at least 3 months; parents/caregivers who could not read and/or write in Portuguese were
excluded.

It was considered as caregiver, the person responsible for most of the care of the patient.
Home mechanical ventilation included invasive ventilation by tracheostomy and non-
invasive ventilation provided at the child’s home.

Complex chronic disease was defined based on the definition of Feudtner: “Any medical
condition that can be reasonably expected to last at least 12 months (unless death
intervenes) and to involve either several different organ systems or 1 organ system
severely enough to require specialty pediatric care and probably some period of
hospitalization in a tertiary care center”(8) and updated according to the ICD-10
diagnostic codes.(9)

The diagnoses were grouped according to the diagnostic group into neuromuscular
disease, respiratory disease, metabolic/neurodegenerative disease, central
hypoventilation and other congenital or genetic defect (which included all the
congenital/genetic defects, except central hypoventilation, which constitutes a different group). They were also grouped according to the trajectory, using the classification of the Association for Children with Life-Threatening or Terminal Conditions and their Families (ACT), which reflects the trajectory of diseases requiring palliative care in a pediatric context. Group 1 represents “life-threatening conditions for which curative treatment may be feasible but can fail”, group 2 covers “conditions where premature death is inevitable, but long periods of intensive treatment are aimed at prolonging life and allowing participation in normal activities”, group 3 includes “progressive conditions without curative treatment options, where treatment is exclusively palliative and may extend over many years” and group 4 represents “irreversible but non-progressive conditions with severe disability and are susceptible to health complications and premature death”.

We used the Pittsburgh Sleep Quality Index (PSQI), validated for the Portuguese population, as the evaluation instrument. After obtaining an informed written consent, caregivers completed the questionnaire anonymously.

The PSQI is a generic instrument that subjectively measures the sleep quality of a subject over a 1-month time interval. It consists of 19 questions, grouped into 7 clinical domains related to sleep quality (subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleep medication, and daytime dysfunction). Each component is evaluated on a scale from 0 to 3. The 7 components are combined to obtain the overall value of sleep quality on a scale from 0 to 21. It helps distinguish between “good sleep quality” and “poor sleep quality”. A global sum of 5 or greater indicates a “poor” sleeper, with major difficulties in at least two components, or moderate difficulties in over three components. An higher overall value reflects a worse sleep quality.

The information obtained was complemented by a questionnaire which included other
variables, such as demographic variables (gender, age, literary abilities, marital status, relationship to the child/adolescent) and clinical variables (presence of sleep disorder) of the caregivers and demographic, clinical and social variables of the child/adolescent (gender, age, diagnosis, attendance of a place of learning quantified in hours, average of daily hours of ventilation and type of interface of ventilation). Ethics Committee provided approval.

Statistical analysis was performed using IBM SPSS Statistics® version 25. The level of significance adopted in the study was 5%.

Results

1. Group of caregivers and patients

PSQI were filled by 29 mothers (87.9%) and 4 fathers (12.1%), with no other type of caregivers included. The sociodemographic details are presented in Table 1. Of the 33 caregivers included in the study, 24.2% suffered from some type of pathology or sleep disorder.

| Sociodemographic details of caregivers |
|---------------------------------------|
| Age (years)                           | 40 (30–53) |
| Academic degree                      | n (%)      |
| Primary school                       | 3 (9.1%)   |
| Middle School                        | 10 (30.3%) |
| High school                          | 13 (39.4%) |
| Bachelor/Master                      | 7 (21.2%)  |
| Marital Status                       | n (%)      |
| Single                               | 3 (9.1%)   |
| Married/Non-marital partnership      | 25 (75.8%) |
| Divorced/Separated                   | 5 (15.2%)  |

Regarding the children/adolescents included in the study, 23 were male (69.7%) and 10 were female (30.3%), with the following age distribution: 6 (18.2%) between 1 and 5 years old; 8 (24.2%) between 6 and 10 years old; 19 (57.6%) between 11 and 18 years old.

Within this group, 81.8% of the children/adolescents attended school daily. The main diagnoses of the patients were categorized into a diagnostic group and an ACT group, as
presented in Table 2.

| Diagnostic group                                    | n (%)        |
|-----------------------------------------------------|--------------|
| Other congenital or genetic defect                  | 15 (45.5%)   |
| Neuromuscular disease                               | 8 (24.2%)    |
| Respiratory disease                                 | 7 (21.2%)    |
| Central hypoventilation                             | 2 (6.1%)     |
| Metabolic/neurodegenerative disease                 | 1 (3%)       |

| ACT group                                           | n (%)        |
|-----------------------------------------------------|--------------|
| 1                                                   | 0 (0%)       |
| 2                                                   | 17 (51.5%)   |
| 3                                                   | 1 (3%)       |
| 4                                                   | 15 (45.5%)   |

The type of interface of ventilation used was face mask in 27 (81.8%) patients, nasal mask in 5 (15.2%) and tracheostomy in 1 (3%). The average of daily hours of home mechanical ventilation was 8 hours (minimum 4 hours; maximum 12 hours), with 48.5% of patients using ventilation for no more than 8 hours, 51.5% between 9 and 15 hours and no patients requiring ventilation for 16 or more hours.

2. Sleep quality

The values obtained in each of the components of the PSQI and its global value are presented in Table 3.
| Component 1 – Subjective sleep quality % |
|------------------------------------------|
| Good 63.6%                               |
| Fairly bad/Very bad 36.4%                |

| Component 2 – Sleep latency % |
|------------------------------|
| Mild to moderate changes 50.0% |
| Few or no changes 50.0%       |

| Component 3 – Sleep duration % |
|-------------------------------|
| > 7 hours 48.4%               |
| 6–7 hours 19.4%               |
| < 6 hours 32.3%               |

| Component 4 – Sleep efficiency % |
|---------------------------------|
| ≥ 85%                           |
| 75–84%                          |
| ≤ 74%                           |

| Component 5 – Sleep disturbance % |
|----------------------------------|
| Mild to moderate 75.0%            |
| Severe 25.0%                      |

| Component 6 – Use of sleep medication % |
|-----------------------------------------|
| No 72.7%                                |
| Yes, once or twice a week 9.1%          |
| Yes, three or more times a week 18.2%   |

| Component 7 – Daytime dysfunction % |
|-------------------------------------|
| Mild 25.0%                          |
| Moderate to severe 75.0%            |

| Mean SD % |
|-----------|
| Global PSQI score 8 4 72.4% |
| Global score ≥ 5 – Poor sleep quality | |
| Global score < 5 – Good sleep quality | 27.3% |

The PSQI global values are presented in Table 4 by diagnostic and ACT group, characteristics of the home mechanical ventilation and attendance of a place of learning.
Table 4
Pittsburgh Sleep Quality Index global values according to patients’ characteristics

| Diagnostic group                                      | Mean PSQI value (SD) |
|-------------------------------------------------------|----------------------|
| Other congenital or genetic defect                    | 6 (4)                |
| Neuromuscular disease                                 | 8 (4)                |
| Respiratory disease                                   | 11 (4)               |
| Central hypoventilation                               | 8 (5)                |
| Metabolic/neurodegenerative disease                   | 7 (NA)               |
| ACT group                                             |                      |
| 2                                                     | 9 (4)                |
| 3                                                     | 7 (NA)               |
| 4                                                     | 7 (4)                |
| Type of interface of home mechanical ventilation      |                      |
| Face mask                                             | 7 (5)                |
| Nasal mask                                            | 7 (4)                |
| Tracheostomy                                          | 9 (NA)               |
| Daily hours of home mechanical ventilation            |                      |
| ≤ 8 hours                                             | 6 (1)                |
| 9-15 hours                                            | 8 (4)                |
| Attendance of a place of learning                     |                      |
| Yes                                                   | 8 (4)                |
| No                                                    | 8 (4)                |

SD: standard deviation; NA: not applicable

There is no statistically significant correlation between the number of hours a day of home mechanical ventilation and the global values of sleep quality ($r=-0.074$, $p = 0.669$).

Nevertheless, it is possible to infer that a greater number of hours of home mechanical ventilation tends to cause a poorer global sleep quality of the caregiver, through a scatter plot (Fig. 1).

The relationship between the global value of sleep quality and the presence of pathology/sleep disorder in the caregiver was studied using extreme diagrams and quartiles (Fig. 2).

Discussion

Main findings of the study

Sleep disturbance is a highly common problem, with multiple contributing factors, and is experienced by family caregivers of children who transition from specialized pediatric facilities to their home.(2) Whereas short-term sporadic exposure to sleep deprivation over the course of a lifetime most likely will not have serious long-term consequences on neural functions, a relatively long and constant period of sleep deprivation, chronic sleep
restriction or fragmentation can lead to serious acute and chronic damage to neural functioning.(10)

The mean global value obtained in the PSQI test for our sample was much higher than that obtained for a healthy normalized Portuguese population, where the mean global score was 1.20 (SD = 0.46), which shows a significantly impaired sleep quality.(11) However, this value is similar to the results of other populations of caregivers of patients with severe psychomotor development (mean global value of PSQI = 8, SD = 4) (12) and caregivers of adolescents with neuromuscular diseases under home mechanical ventilation (mean global value of PSQI = 7.4, SD = 4.7).(13)

Most of the sample (72.4%) obtained global scores greater than or equal to 5, which indicates the presence of difficulties in, at least, two components of the questionnaire, showing a higher risk for sleep disturbance.(11) On one hand, the literature associates sleep disruption with short-term health consequences (heightened stress response, pain, depression, anxiety and cognition, memory and performance deficits) and long-term health consequences for the caregivers (hypertension, cardiovascular disease, weight gain, metabolic syndrome and type 2 diabetes mellitus).(5) On the other hand, sleep disruption is associated with hampering the activities and duties of the caregiver, as well as the patient care.(14)

More than half of the caregivers (63.6%) perceived their own quality of sleep as good, despite the global value obtained in the PSQI test indicated a "good sleep quality" in only 27.3%. This may be because the caregiver gets accustomed to it.(13)

Caregivers of children/adolescents with respiratory disease were the ones that had the worst values in sleep quality (mean PSQI value = 11), followed by neuromuscular disease and central hypoventilation (mean PSQI value = 8). In previous studies, caregivers of children ventilated due to respiratory pathology presented mean PSQI values of 8.69,(14)
Caregivers of young people with neuromuscular diseases under home mechanical ventilation values of 7.4 (13) and mothers of children with congenital central hypoventilation syndrome values of 7.5.(15) There is an accentuated difference in the mean PSQI value in the respiratory group when compared to our study.

Caregivers of ACT group 2 presented the worst sleep quality (mean PSQI value = 9). Although no reports have been found in the literature that make this comparison between ACT groups, we can hypothesize an association to the longer trajectory of disease.

Regarding sleep duration, approximately half of the sample had an average sleep time superior to 7 hours, but 32.3% slept 6 hours or less per day. The basic need for sleep in a healthy adult is roughly 7.5 to 8.5 hours,(10) and less than 6 hours of sleep per night is not recommended, because deficits in cognitive functioning, especially memory, and response times have been found.(16)

Half of our sample had sleep efficiency indices below 85% (the normal value is 80%), which may be associated with drowsiness and diurnal dysfunction. A study has shown a significant difference in sleep efficiency between the caregivers of ventilated children and the caregivers of healthy children.(17)

A very significant percentage of caregivers reported sleep disturbance and/or daytime dysfunction, although this may not be directly related to the need for care provided to the child/adolescent. It was described that 52% of caregivers of ventilated children reported waking at least once a week to perform nighttime caregiving duties compared with 9% of caregivers of healthy children.(17) Furthermore, in families of technology-dependent children, a large proportion of families (22 out of 36) experienced regular sleep disruption, where parents reported getting up in the night at least twice a week.(18)

Approximately 25% of the caregivers need sleep medication, at least once a week, which is similar to what is recorded for a population of caregivers of ventilated children (24.1%).
Sleep quality of the caregivers didn’t vary significantly with the attendance of a place of learning. In a study of children with respiratory disease, no differences in primary sleep outcomes were found between caregivers who completed questionnaires during the school year versus the summer/holidays. (14) The type of interface and the number of daily hours using home mechanical ventilation didn’t seem to interfere with sleep quality. The duration of home mechanical ventilation and the time of the day when it was performed were identified as important factors for sleep disruption, with a greater negative impact with the increase in daily hours of home mechanical ventilation. (18) However, the longer-term use of non-invasive ventilation appears to result in an adjustment by the caregiver, with consequent improvement in sleep quality. (19)

Strength and limitations of the study

Within the limitations of the study, it is to highlight that the sample size is relatively small, making it difficult to find significant associations. PSQI doesn’t provide detailed information on specific factors, which may vary depending on the underlying pathology and, therefore may influence the sleep quality of caregivers. The study was based on a subjective self-completion tool that can lead the caregiver to underestimate/overestimate the quality of the measured parameters. The fact that about 25% of the sample presented a sleep disorder may constitute a bias to the results found. Sleep may be influenced by other variables not considered in this study (environmental issues, sleep hygiene habits, presence of one or more caregivers, child/adolescent’s disease stage).

It would be useful to carry out a prospective and longitudinal study, which would allow the questionnaires to be applied immediately before the onset of home mechanical ventilation.
and after a period of 3 to 6 months, in order to determine the impact of home mechanical ventilation on the sleep quality of the caregivers, reducing the interference of other external factors.

**What this study adds**

This study is one of the first in the international literature to specifically assess sleep quality of caregivers of children/adolescents with complex chronic disease under home mechanical ventilation. The sleep quality of the caregivers of children/adolescents under home mechanical ventilation is lower than that recorded in the healthy Portuguese population.

The authors concluded that the caregivers of children/adolescents with respiratory disease and of ACT group 2 had the worst values in sleep quality (mean PSQI value = 11 and 9, respectively). There is an accentuated difference in the mean PSQI value in the respiratory group of other studies when compared to our study. Besides, we haven’t found previous literature comparing sleep quality of caregivers of different ACT groups.

More than half of the caregivers perceived their own quality of sleep as good, despite the global value obtained in the PSQI test indicating a "good sleep quality" in less than one third.

**Conclusion**

Sleep quality of caregivers of children/adolescents under home mechanical ventilation is lower than that recorded in the healthy Portuguese population and presents values similar to those described in particular groups in the literature.

Given the data obtained, the authors consider that it is essential to sensitize health professionals to assess sleep quality of the caregivers of patients with complex chronic disease, given the relevance, not only in performing of their duties, but also in their own
health and quality of life. Therefore, taking care of the caregivers should be one of the axes of the medical intervention.

Declarations

Authorship

Raquel Gonçalves made a substantial contribution to the concept and design of the work, contributed to acquisition, analysis and interpretation of data, drafted the article and critically revised it for important intellectual content.

Cristiana Martins made a substantial contribution to the concept and design of the work, and contributed to acquisition, analysis and interpretation of data.

Ana Borges contributed to acquisition of data.

Núria Madureira made a substantial contribution to the concept and design of the work, and critically revised it for important intellectual content.

Cândida Cancelinha made a substantial contribution to the concept and design of the work, contributed to acquisition, analysis and interpretation of data, drafted the article and critically revised it for important intellectual content and approved the version to be published.

All authors gave final approval.

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Research ethics and patient consent: Written consent was obtained from the patients’ parents for this information to appear in a journal article. Ethics committee (Comissão de Ética para a Saúde do CHUC) provided approval (number CHUC-125-18).

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Figures
Figure 1

Scatter plot between the number of hours a day of home mechanical ventilation and the global values of Pittsburgh Sleep Quality Index
Figure 2

Presence of pathology/sleep disorder in the caregiver and the global value of Pittsburgh Sleep Quality Index: No – average: 7 (SD=4); Yes – average: 9 (SD=4)

Supplementary Files

This is a list of supplementary files associated with the primary manuscript. Click to download.

Appendix.docx