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Sleep quality in children requiring palliative care and their families

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Summary. Aim. Sleep disturbances are common symptoms between children with life-threatening (LTDs) and life-limiting diseases (LLDs), impacting their parents' sleep quality. Nevertheless, up-to-date exhaustive data regarding sleep problems in children with LTD and LLDs, particularly related to their clinical condition, are poor, making it difficult to implement supportive measures. This study aims to describe the sleep quality of chronically ill children, along with that of their parents. In addition, the correlation between the child's sleep quality and the burden of children's care was investigated. **Methods.** This is a cross-sectional study conducted among parents of children and adolescents in care at our regional center for pediatric palliative care (PPC) and pain control, in the period from August 2020 to September 2020. **Results.** The need to change posture and pain are mostly triggers of a high number of arousals for chronically ill children, increasing the need for nightly procedures and disrupting children's and parent's sleep. **Conclusion**. Our study underlines the importance for families of children with LTD and LLDs to receive appropriate support from a PPC team to manage sleep disturbances, paying more attention to the evaluation and the quality of children's sleep.

Key words. Life-limiting conditions, life-threatening conditions, complex healthcare needs, pediatric palliative care, sleep quality.

Valutazione della qualità del sonno dei bambini in cure palliative e delle loro famiglie.

Riassunto. Obiettivi. I disturbi del sonno sono un sintomo comune tra i bambini affetti da malattie potenzialmente letali (LTD) e limitanti la vita (LLD). Tali disturbi influiscono anche sulla qualità del sonno dei loro genitori. Tuttavia, dati aggiornati e esaustivi sui problemi del sonno nei bambini con LTD e LLD, in particolare legati alla loro condizione clinica, sono scarsi e questo rende difficile l'implementazione di opportune misure di supporto. L'obiettivo di questo studio è quello di descrivere la qualità del sonno dei bambini affetti da LTD e LLD e quella dei loro genitori. Inoltre, è stata studiata la correlazione tra la qualità del sonno del bambino e il suo carico di malattia. **Metodi.** Lo studio è stato condotto tra i genitori dei bambini e adolescenti in cura presso il Centro regionale veneto per le cure palliative pediatriche (PPC) e il controllo del dolore, tra agosto 2020 e settembre 2020. **Risultati.** La necessità di cambiare posizione e il dolore sono i fattori che principalmente causano un elevato numero di risvegli nei bambini, aumentando la necessità di eseguire procedure notturne e disturbando di conseguenza il sonno sia dei bambini che dei genitori. **Conclusioni.** Lo studio sottolinea l'importanza per le famiglie dei bambini affetti da LTD e LLD di ricevere un supporto adeguato da parte di un team esperto in PPC, prestando maggiore attenzione alla valutazione e alla qualità del sonno dei bambini.

Parole chiave. Patologie limitanti la vita, patologie pericolose per la vita, esigenze sanitarie complesse, cure palliative pediatriche, qualità del sonno.

Introduction

Over the past decades, science and technology have led to an increase in the survival of many children suffering from life-threatening diseases (LTDs) and life-limiting diseases (LLDs). As a consequence, a steadily growing number of children live with complex healthcare needs throughout their lives and need constant support and long-term care in the pediatric palliative care (PPC) setting. PPC is provided to improve the global well-being of both children and their families providing adequate assistance for clinical, psychosocial, and spiritual issues[1-4].

Different papers have shown that sleep insufficiency in children who suffer from LTDs and LLDs is a very common problem that ranges from 50 to 80% of cases.[5-7] Sleep disturbances have a negative impact not only on the child but also on the global quality of life (QoL) of their families[8-11]. Of note, according to a study by Meltzer et al., sleep disruptions are also associated with higher rates of depression and anxiety in parents of children with complex healthcare needs[12].

Consequently, the identification and characterization of children's sleep problems result to be very important issues in the context of PPC, to provide children and families with proper supporting measures

able to prevent poor social, emotional and cognitive outcomes and to improve their QoL[13].

To this aim, different studies tried to define clinical instruments able to measure sleep quality both in healthy children or in neurologically impaired children or with LLCs[14-16].

Nevertheless, quality of sleep is a complex phenomenon that includes quantitative (i.e., sleep duration, sleep latency or number of arousals) and subjective (i.e., "depth" or "restfulness") aspects, making it difficult to define and characterize objectively. The available instruments to measure sleep quality, even when tailored to children with complex clinical needs, do not consider the individual features that characterize the children's diagnoses[16].

Consequently, up-to-date exhaustive data and information regarding sleep problems in children with LTD and LLDs, particularly concerning their clinical condition, are poor.

This study aims to investigate and describe the sleep quality of chronically ill children in care at our regional center for PPC, along with that of their parents. In addition, the correlation between the child's sleep quality and the burden of children's care (such as the use of medical devices, the need for overnight procedures) was investigated.

Patients and methods

Study design, participants and setting

This is a cross-sectional study conducted among parents of children and adolescents in care at our regional center for PPC and pain control, in the period from August 2020 to September 2020. The PPC center of Padua is in charge of about 180 patients a day, providing high-level assistance at home and as an in-hospital pediatric hospice. The PPC network is composed of a multidisciplinary team of palliative care specialists who coordinates the healthcare providers and manages the different complex needs of patients and their families in different familiar backgrounds. Exclusion criteria considered were the decline to participate in the study, a poor understanding of the Italian language and the registration to the PPC network after the questionnaire formulation (30/03/2020). Informed consent was obtained from all participants.

Cross-sectional survey

A descriptive observational survey was carried out using a 9-item *ad hoc* parental-reported sleep questionnaire, formulated according to the existing literature on sleep quality in healthy and disabled children (PubMed, Cochrane and Cinahl database set). The questions concern parents' sleep quality and the perception about the quality of sleep of their children during the previous 4 weeks [Appendix 1].

Furthermore, the questionnaire investigated the time of sleep, the wake time, the place where children used to sleep, the use of sleeping drugs, the number and duration of arousals. The survey also assesses the principal reasons for arousals. For this question, it was possible to choose multiple answers (e.g., pain or crying, epileptic seizures, passive changing position and other factors correlated with the environment). Another question investigates the procedures that most frequently could disrupt children's sleep (multiple answers question).

The last two questions, adapted from the SNAKE questionnaire,[16] investigated children's and caregivers' perceived sleep quality through an overall rating divided into "very good," "good," "very poor," "poor," and "satisfactory."

Demographic data (i.e., gender, age, nationality) and additional data regarding patients' disability and care needs (i.e., number and type aids for each patient, physiotherapy sessions) were also collected.

The interviews were conducted with one of the parents by phone or during home visits by the team of PPC specialists.

A period of clinical stability was considered for the assessment of sleep quality. The quality of sleep at conditions of acute problems (e.g., ongoing infections, hospital admissions) was not evaluated.

Data analysis

Mean, median, rate and range were used to describe variables. Data analysis was performed with the statistical software R (free software under GNU-GPL license). When available, correlations between numerical and categorical variables were calculated with ANOVA test, between numerical variables with Pearson's test, and between categorical variables with Chi-square analysis, or Fisher's exact test.

Results

Study population

During the study period, 171 children and adolescents were in care at our PPC center. In total, 29 children had to be excluded due to the following reasons: enrollment after 30 March 2020 (n=15), decline to participate (n=7), and poor Italian language (n=7). The final sample therefore composed of 142 patients. They were divided into three groups accordingly to their diagnosis: oncological (n=9, 6%), non-oncolog-

ical (n=123, 87%) and undiagnosed conditions requiring PPC (n=10, 7%). The non-oncological group of patients includes different pathologies, such as neuromuscular disorders, metabolic diseases, genetic or chromosomal diseases, and neuropathies or neurological lesions. Characteristics of participating children are summarized in table 1.

Survey results

Most children sleep in the same room as their parents (41%, n=58) and the same bed for 16% of them (n=23). The use of sleeping drugs regards 23% of the sample (n=33), but the quality of sleep is considered "poor" or "very poor" in 50% of this group. A total of 43% of the sample declared one or two arousals per night (n=61), and 30% declared three or more awakenings (n=43). Most children and teens woke up on their own during the night (71%, n=101), mostly due to the need to change position (55%, n=58). The most common caregiver assistance maneuvers were the change of position (66%, n=42) and the pain management (18%, n=12). Parents mostly perceived the quality of the children's sleep as good or very good (65%, n=93), while their own sleep was perceived primarily as negative (40%, n=57). The overall survey results are presented in table 2.

Table 1. Characteristics of included patients (n=142).

Characteristics	n (%)
Males	66 (46)
Age (years), mean (SD)	10 (6)
Nationality:	
■ Italian	110 (77)
■ Other European countries	10 (7)
■ African countries	11 (8)
■ Asian countries	8 (6)
■ Brazil	2 (1)
■ USA	1 (1)
Physiotherapy:	
■ Weekly (performed by a specialist)	84 (61)
■ No physiotherapy/passive exercise	54 (39)
Medical devices: *	
■ No medical devices	33 (24)
■ One device	64 (46)
■ Two devices	33 (24)
■ Three devices	8 (6)
■ Four devices	1 (1)
Most common devices:	
■ Gastrostomy	67 (48)
■ Ventilator	60 (43)

*Ventilator, tracheostomy, gastrostomy tube, central venous access.

Table 2. Sleep-related issue (n=142).

Q	Issue	n (%)
Q1	Wake-up time, mean:	7.45 am
	■ Wake-up time range	5.30 am – 11 am
Q2	Sleeping time, mean:	10.30 pm
	■ Sleeping time range	8.30 pm-3 am
	Hours of sleep, mean	9 hours
Q3	Place of sleep:	
	■ Bedroom with parents	58 (41)
	■ Parents' bed	23 (16)
	■ Own room	49 (34)
	■ Shared room with siblings	12 (9)
Q4	Sleeping drugs:	
	■ No drugs at all	109 (77)
	■ Melatonin	26 (18)
	■ Benzodiazepines	7 (5)
Q5	Total arousals:	
	■ No awakenings	38 (27)
	■ One awakening	20 (14)
	■ Two awakenings	41 (29)
	■ From three to six	43 (30)
Q6	Autonomous arousals:	101 (71)
	■ Constant sleep*	41 (28.87)
	Cause of arousal**:	
	■ Pain or crying	15 (14)
	■ Epileptic seizures	9 (9)
	■ Passive changing position	58 (55)
	■ Others***	55 (53)
Q7	Nightly procedures:	
	■ Yes	64 (42)
	– But children do not wake up	5 (8)
	■ No	78 (55)
	Type of maneuvers:	
	■ Posturing	42 (66)
	■ Pain management	12 (18)
	■ Tracheal aspiration	10 (16)
Q8	Children's quality of sleep:	
	■ Very good	33 (23)
	■ Good	60 (42)
	■ Quite satisfactory	41 (29)
	■ Bad	7 (5)
	■ Very bad	1 (1)
Q9	Caregivers' quality of sleep:	
	■ Very good	8 (6)
	■ Good	36 (25)
	■ Quite satisfactory	41 (29)
	■ Bad	45 (32)
	■ Very bad	12 (8)

*Some children undergo posture change or mobilization procedures that do not involve autonomous arousal.
** Parents could choose more than one cause of arousal only when undecided.
***Including light, TV running and noise in child's bedroom (environment factors).

Correlation analyses

The child's sleep quality correlated with the number of arousals (p<0.001) and the use of pharmacological treatment (p=0.041). The children's and caregivers' sleep quality correlated significantly (p <0.0001) (table 3), and the parent's sleep quality is significantly improved with the increasing age of their child (p=0.019).

The parent's sleep quality does not correlate with the children's place of sleep, while it worsened significantly in relation to the number of child awakenings (p<0.001) (table 4). The number of arousals did not correlate with the type of diagnosis, the type of physiotherapy procedures or the procedures performed during the night (table S1).

Discussion

Sleep disturbances are one of the most common symptoms between children with LTDs and LLDs, also impacting the sleep quality of parents who provide nightly care[16].

Sleep quality complaints in chronically ill children can be related to many different factors, such as medical care, pain, psychiatric conditions, genetic pathologies, or neurologic and metabolic diseases[5]. Furthermore, children and adolescents' sleep problems can express emotional disturbances, physical diseases, and school difficulties[17].

In our sample, children's and parents' sleep quality were significantly correlated. Parents' sleep quality is significantly improved with the increasing age of their child (p=0.019) and worsened significantly in relation to the number of child awakenings (p<0.001). Arousals were mainly caused by the need to change position and by pain. For instance, all patients in care at the center are prescribed acute pain treatments and therapies to manage chronic pain.

Of note, stratifying patients accordingly to the use of sleeping drugs, a trend to a better sleep quality was observed for non-treated children. The reason behind this finding is not clear. It may be related to both a global worse clinical condition of treated children and to a reduced effect of the administered therapies. For instance, melatonin is widely used in children with neurodevelopmental disabilities, but there are

Table 3. Children's sleep quality correlations.

	Sleep rating					p-value
	Very poor	Poor	Satisfying	Good	Very good	
Arousals, mean (SD)	5.00 (0.00)	3.14 (1.68)	2.49 (1.34)	1.62 (1.35)	0.88 (1.02)	<0.001
Medical devices, mean (SD)	2.00 (0.00)	1.14 (0.90)	1.38 (0.95)	1.09 (0.85)	0.94 (0.75)	0.208
Sleeping drugs, n (%):						
■ Yes	1 (3)	2 (6)	14 (42)	13 (39)	3 (9)	0.041
■ No	0 (0)	5 (4)	27 (25)	47 (43)	30 (27)	0.030
Caregivers' quality of sleep, rating:						
■ Very poor	1	1	4	4	2	
■ Poor	0	5	20	15	5	
■ Satisfying	0	1	13	19	8	<0.0001
■ Good	0	0	4	22	10	
■ Very good	0	0	0	0	8	

Table 4. Parent's sleep quality correlations

	Sleep rating					p-value
	Very poor	Poor	Satisfying	Good	Very good	
Place of sleep, n (%):						
■ Parents' bed	4 (7)	13 (22)	20 (34)	17 (29)	4 (7)	
■ Shared room with siblings	0 (0)	3 (25)	5 (42)	4 (33)	0 (0)	0.495
■ Own room	5 (10)	19 (39)	13 (26)	9 (18)	3 (6)	
■ Parents' bed	3 (13)	10 (43)	3 (13)	6 (26)	1 (4)	
■ Child's arousals, mean (SD)	2.17 (1.47)	2.31 (1.44)	1.83 (1.24)	1.36 (1.53)	0.12 (0.35)	<0.0001

no clinical guidelines addressed to its prescription and effects monitoring in such patients[9,18].

Our study shows that caring for a family with children harboring LTDs and LLDs may require many procedures during the night, resulting in important sleep disruption. The difficulty of establishing some guidelines to manage sleep disturbances lies in the great variety of diagnoses. Therefore, it is important to monitor the sleep and well-being of patients and their parents by the specialist PCC team to improve the QoL of the whole family. In this context, the collaboration between an interdisciplinary pediatric sleep clinic and a PPC specialist could improve specific interventions and provide support for the families to improve their sleep quality and quantity[19].

Conclusion

The need to change posture and pain are mostly triggers of a high number of arousals for chronically ill children, increasing the need for nightly procedures and disrupting children's and parent's sleep.

In the literature, there are no guidelines about the best approach to managing these situations and improving the quality of sleep for children and their families. This suggests that sleep disturbances in children requiring PPC have received limited attention. However, it has been well demonstrated that children and their families could achieve a better QoL by reducing sleep problems.

Our study underlines the importance for these families to receive appropriate support by a PPC team to deal with the management of having a child with life-limiting or threatening conditions also during sleep hours, paying more attention to the evaluation and the quality of children' sleep. The collaboration between an interdisciplinary pediatric sleep clinic and a PPC specialist could be of great help to define specific interventions.

Conflict of interests: the authors have no conflict of interests to declare.

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