HK J Paediatr (new series) 2020;**25**:89-97

Original Article

The Risk Factors Associated with Sleep-related Problems in Children with Profound Intellectual Disability

CK ***Chow***, SN ***Wong***, LCK ***Ma***, GPG ***Fung***, WL ***Yam***, HB ***Chan***

Abstract ***Objective:*** To estimate the prevalence and identify the risk factors of different sleep disorders, in a local sample of children with profound intellectual disability. ***Methods:*** The study was conducted in students with profound intellectual disabilities in a special school. Each student was evaluated by the Chinese version of Children's Sleep Habits Questionnaire by their parents. Other medical information was collected and analysed with the score of the questionnaire. ***Results:*** Data from 67 participants were included in the analysis. 84% of the subjects were considered as having sleep disorders. Children with epilepsy had a significantly higher total sleep score compared to children without epilepsy (49.66±7.53 vs 45.27±6.87, p=0.02). Non-boarding students and those with medical problems also had significantly higher scores. Logistic regression analysis showed that epilepsy was significantly associated with sleep disorder (adjusted odd ratio 7.99, p=0.02). Epilepsy was associated with higher sub-scores for bedtime resistance, sleep onset delay and sleep-disordered breathing. Obesity was associated with higher sub-score for sleep-disordered breathing. ***Conclusions:*** Sleep disorders were common among children with profound intellectual disabilities, and epilepsy was significantly associated with sleep disorder in this group of children. Correct identification of the sleep disorders, followed by provision of different sleep interventions are recommended.

Key words Cerebral palsy; Epilepsy; Intellectual disability; Sleep-related problem

Introduction

Intellectual disability (ID) is a lifelong neurodevelopmental disorder, characterised by both intellectual and adaptive functioning deficits in conceptual, social and practical domains.[1] In addition to cognitive impairment, students with ID also face many health-related problems, including motor deficits, epilepsy, sleep disturbance, mental illness, vision and hearing impairment.[2]

The classification of ID into different grades of severity, which was previously defined by the intelligence quotient (IQ), is now emphasized on the adaptive functioning of the subject.[1] A "three-tier system" is still normally used in Hong Kong for operational classification. Children are classified into "mild", "moderate" and "severe" grades for the purpose of special education and training services.[3] For example, the "severe" grade in Hong Kong corresponds to "profound" grade in the previous World Health Organisation (WHO) classification system. Therefore, a "school for children with severe ID" in Hong Kong is receiving students with IQ less than 20 (results in severe limitation in self-care, continence, communication and mobility).[4] In Hong Kong, the prevalence rate of ID is

Department of Paediatrics and Adolescent Medicine, United Christian Hospital, 130 Hip Wo Street, Kwun Tong, Kowloon, Hong Kong SAR, China

CK **Chow** (周哲光) *FHKCPaed, FHKAM(Paed)*
SN **Wong** (王雪娜) *FHKCPaed, FHKAM(Paed)*
LCK **Ma** (馬子軍) *FHKCPaed, FHKAM(Paed)*
GPG **Fung** (馮寶姿) *FHKCPaed, FHKAM(Paed)*
WL **Yam** (任惠蓮) *BSN & MSc(Nursing), FHKAN(Paed)*
HB **Chan** (陳衍標) *FRCPCH, FHKAM(Paed)*

Correspondence to: Dr CK **Chow**
Email: cck904@ha.org.hk

Received January 2, 2018

estimated to be 1.0% to 1.4%[5] and in this population, about 85% are within mild grade, 10% are within moderate grade, and the rest are within the severe grade range.[3] In 2015, 679 students were enrolled in a school for children with severe ID, in contrast with over 300,000 enrolments in ordinary primary schools.[6]

Sleep disorders can be classified according to different classification systems, but in general, they can be divided into two major categories: dyssomnias and parasomnias. Dyssomnias refer to those involving initiation or maintenance of sleep, or excessive sleepiness. Parasomnias refer to abnormal events that disrupt sleep after its initiation.[7,8] The International Classification of Sleep Disorders (third edition, ICSD-3), developed by American Sleep Disorders Association, is one of the most comprehensive classifications available currently. The ICSD-3 identified seven major categories that include insomnia disorders, sleep-related breathing disorders, central disorders of hypersomnolence, circadian rhythm sleep-wake disorders, sleep-related movement disorders, parasomnias, and other sleep disorders.[9] In this classification system, for example, paediatric obstructive sleep apnoea belongs to the category of sleep-related breathing disorders, which its clinical findings (such as snoring, laboured or obstructed breathing, daytime consequences) and polysomnographic criterion have been described. On the other hand, the parasomnias are divided into three groups, namely non-rapid eye movement (such as confusional arousal, sleep walking, sleep terrors), rapid eye movement (such as nightmares) and others.[9]

Sleep disorders in children with ID are common yet under-recognised problems. They gain relatively little attention from parents, clinicians and researchers as compared with other clinical agendas. Parents rarely voice out the problem until it becomes long term and affects the whole family. Clinicians, however, have tended to regard the use of medication as the only key to this problem.[7,10] Nonetheless, this problem is often a significant source of parental stress.[11,12] The estimated prevalence of children with ID having sleep disorders vary from 13% to 86%, depending on case definitions.[13-15] For example, a questionnaire-based study assessed the prevalence of sleep problems in children with ID and found that 8.6%, 14.8%, 27.9% and 35.3% of children with mild, moderate, severe and profound ID had severe sleep problems respectively.[16] It was also noticed that there were higher proportions of dyssomnias, including obstructive sleep apnoea, and parasomnias in children studying in special schools.[7]

As mentioned, children with ID have different co-morbidities, and the associations between these and sleep disorders have been explored in previous studies. There is often a bi-directional association, in which these co-morbidities have a negative impact on their sleep, and in turn the sleep disorders can potentiate the co-morbidities.[12,17-24] For the aforementioned questionnaire-based study, it assessed children with ID from mild to profound range and concluded that children with severe sleep problems had more severe levels of ID, had greater frequencies of epilepsy and cerebral palsy than those without severe sleep problems. Besides, children with severe sleep problems showed more daytime behavioural problems, such as aggression and hyperactivity.[16] These findings corroborate the results of other epidemiological studies. A study using a standardised observation protocol studied subjects with ID living in a rehabilitation centre, and it concluded that severe locomotor disability and active epilepsy were independent predictors of increased daytime sleep and wake-sleep transitions.[25] A study recruited children with ID and epilepsy and found that about one-third of them had obstructive sleep apnoeas and periodic limb movements.[26] A study also demonstrated the association between sleep problems and anxiety, in which higher levels of sleep problems are associated with higher levels of anxiety in children with ID.[27] Therefore, it is important to identify these co-morbidities as potential risk factors for sleep disorders in this group of children, as interventions targeted at the co-morbidities could potentially improve the sleep disorders and vice versa.

Local data on the prevalence of sleep disorders in children with ID is lacking, but an understanding of this important issue may help improve planning and management of this group of children. Few studies have examined the risk factors of sleep disorders in children with profound ID, especially in a special school setting. They are the ones who have more complex medical background but receive relatively little attention from society and researchers. The aim of this study was to estimate the prevalence and identify the risk factors of different sleep disorders in a local sample of children with profound ID studying in a special school.

Methods

Recruitment of Subjects

We recruited all the families with children studying in Haven of Hope Sunnyside School in the academic year

2015/16 as the study population (82 families in total). This special school is one of the 10 "schools for children with severe ID" in Hong Kong, for children from six to 18 years old. As discussed above, its students were all classified as profound ID (IQ less than 20, according to previous WHO classification system) in the pre-school assessment before they entered this school. This school also provides boarding services for the students at the discretion of their families, but all of them will return home in weekends and long holidays. All the students in this school are under the Integrated Care and Community Support (ICCS) programme, which medical staff in a regional hospital are providing medical assessment, visits and opinion to the students and the school staff.

Data Collection

As part of the ICCS programme, all the families of the students in this school received an invitation letter to fill in a sleep questionnaire (see "materials" below). This questionnaire was distributed by the school staff and the process was co-ordinated by the ICCS nurse in the school. Besides the results of the questionnaire, other information of the students, including demographic information and co-morbidities was collected from the students' medical profile, and all data was entered in a standardised data collection form without any personal identification.

In particular, demographic information including age, sex, boarding status, latest body weight and height was provided by the school in the students' school record. The co-morbidities collected were epilepsy, cerebral palsy, visual impairment and psychiatric disease. The use of medication and mobility status (walk unaided, walk with assistance, wheelchair-bound or bed-bound) were also included. The medical diagnoses of the co-morbidities were all reviewed by the medical team, so as to ensure they were appropriate and most updated.

Subjects would be excluded if the family could not provide any information about the children's sleeping habit, or the family did not consent to use children's data for research purpose. Written approval from the school and local ethics committee was obtained for this study.

Materials

Each participant's sleep habit was evaluated with the Chinese version of the Children's Sleep Habits Questionnaire (CSHQ). This questionnaire is a screening instrument designed for surveying sleep habits and sleep disturbances, and identifying both behaviourally-based and medically-based sleep problems in school-aged children.[28] It was seen as acceptable for use with older children and those with developmental delay.[27,29] It is a parent-reported questionnaire consisting of 50 questions, in which 33 of them are scored, and it takes around 15-20 minutes for the family to complete the whole questionnaire. Each question enquires about an item of the child's usual sleeping habit, and is scored as 3 if this item occurs usually (5-7 times per week), 2 if it occurs sometimes (2-4 times per week) or 1 if it occurs rarely (0-1 times per week). The 33 scored items can be divided into eight subscales, namely bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep-disordered breathing and daytime sleepiness. Each completed questionnaire will yield eight sub-scores and a total score. Therefore, this questionnaire is a comprehensive tool for assessing different types of sleep disorders, including dyssomnias and parasomnias. Other non-scored items allow the assessors to gather more information about the children's sleeping habits. Families were instructed to consider the child's usual sleeping habit (preferably in past 1 week and free of recent deviation from the norm such as illness or change of environment).

CSHQ has been validated and showed adequate internal consistency for both the community and clinical sample. A cut off total score of 41 yielded a sensitivity of 0.80 and specificity of 0.72.[28] The Chinese version of CSHQ has been developed, and shows good reliability, content validity and construct validity.[30]

A high CSHQ score warrants further medical attention, which the correct diagnosis of sleep disorders may require further information. For example, detailed history especially complaints from the care-taker, developmental and family history, sleep diary and physical examination may be required to establish a diagnosis.

Statistical Analysis

Statistical analysis was performed using Statistical Package for the Social Sciences software (IBM SPSS Statistics for Windows, Version 23.0. Armonk, NY: IBM Corp.). Categorical data were compared using the Chi-square test or Fisher Exact test (for cells less than 5), and odds ratio (OR) with 95% confidence interval (CI) was calculated. Continuous variables were compared using the independent t test or Mann-Whitney U test. Multivariate logistic regression was used to determine the effect of individual risk factors on outcome. A two-sided p-value of ≤ 0.05 was considered significant.

Results

Eighty-two families were invited to fill in the sleep questionnaire. Sixty-seven completed questionnaires were received which constituted a response rate of 82%. None of the subjects fulfilled the exclusion criteria as stated above. The mean CSHQ total score of all the subjects was 48.3±7.6. The demographics and characteristics of the subjects are summarised in Table 1.

In this study, we investigated possible risk factors that may affect sleep in these children. Comparison of total sleep score between children with or without risk factors is shown in Table 2. Children with epilepsy had a significantly higher total sleep score compared to children without epilepsy (49.66±7.53 vs 45.27±6.87, p=0.02). Non-boarding students also had a significantly higher score, as well as presence of medical problem. Other factors including adolescent age group ≥13 years old (49.00±7.94 vs 47.68±6.91) and obesity (51.71±7.89 vs 47.88±7.49) showed a trend towards higher score, but did not reach statistical significance. Presence of cerebral palsy, psychiatric disorder or mobility status was not significantly associated with higher scores.

If a total score of 41 is used as cut-off value for sleep disorder, 56 children in this study were defined as having sleep disorder and were included into the "Study Group". Eleven children had total score less than 41 and were included in the "Comparison Group". Comparison of risk factors for sleep disorder (Table 3) showed that epilepsy (OR 4.78, 95% CI 1.22-18.7, p=0.02), as well as presence of medical problem (OR 5.83, 95% CI 1.26-27.02, p=0.02) were associated with significantly higher risk of sleep disorder.

Logistic regression analysis comparing the two groups (Table 4) showed that, after elimination of confounding

Table 1 Demographics and characteristics of subjects

	n (%)
Total number of subjects	67 (100)
Male	37 (55)
Female	30 (45)
Age (year)	14±4.7
Boarding student	38 (57)
Obesity (Body mass index >25)	7 (10)
Epilepsy	
No	22 (33)
Yes. Not on AED	4 (6)
Yes. On one AED	21 (31)
Yes. On two or more AEDs	20 (30)
Cerebral palsy	28 (42)
Mobility status	
Walk unaided or with assistance	32 (48)
Wheelchair-bound or bed-bound	35 (52)
Visual impairment	11 (16)
Psychiatric disease	5 (7)

AED = Anti-epileptic drug

Table 2 Comparison of total sleep scores between different groups

	Total sleep score		
	Condition present	Condition absent	p-value
Male gender	48.05±8.11	48.57±6.98	0.79
Adolescent age group	49.00±7.94	47.68±6.91	0.49
Epilepsy	49.66±7.53	45.27±6.87	0.02*
Cerebral palsy	47.46±7.49	48.87±7.67	0.46
Bed or wheelchair bound	49.23±7.97	47.25±7.09	0.29
Psychiatric problem	45.80±8.23	48.48±7.55	0.45
Live in hostel	46.29±7.51	50.90±6.94	0.01*
Obesity	51.71±7.89	47.88± 7.49	0.21
Presence of medical problem	49.10±7.41	43.00±6.71	0.02*

* p-value ≤0.05

factors, epilepsy was significantly associated with sleep disorder (adjusted OR 7.99, 95% CI 1.48 - 43.25, p=0.02).

Further analysis of sub-scores for risk factors for sleep problems (Table 5) showed that different risk factors were associated with higher sub-scores for different categories. Epilepsy was associated with higher scores for subscales 1, 2 and 7 (bedtime resistance, sleep onset delay and sleep-disordered breathing respectively). Obesity was associated with higher scores for subscale 7 (sleep-disordered breathing).

Discussion

Sleep Disorders and ID

The result of our study corroborates those of other epidemiological studies, showing a high prevalence of sleep disorders in children with ID. Using the cut-off point of 41, which was also used in other similar studies,[21,27] 84% of the subjects were considered as having sleep disorders. Prevalence rate of up to 77%[27] and 86%[14] have been reported. We believe the wide range of prevalence rates could be attributed to different methodology, clinical definition and sample group. We focused on children with profound ID while some were studying a heterogeneous group of children with different levels of ID. A higher rate of sleep disorders would be expected in students studying in special school with more severe ID.

The biological basis of sleep disorders in children with ID had been described in the literature. These children may have altered perception of different environment clues (such as light-dark cycle, food schedule) and also show endogenous dysfunction in hormonal release. These factors are important in establishing a synchronised circadian rhythm, so it will potentially affect the development of a normal sleep-wake cycle. They may present with abnormal sleep architecture as seen in polysomnographic study, such as abnormal duration of the sleep cycle.[31]

Despite the high prevalence rate of sleep disorder in this group of children, we notice that this problem receives relatively little attention in society and amongst medical practitioners. It was described as "a neglected topic", which medical students, nurses, clinical psychologists and even paediatric postgraduates receive very little teaching and training.[12] Medical practitioners seldom enquire about this, partly because they may be overwhelmed by other topics considered to be more important. Some parents themselves may also hesitate to seek help as they may consider this problem a chronic one and thus become part of the "normal life" of these children.[10] Even when this problem is brought to the medical practitioners, they are often offered sedatives which do not help, and sometimes even worsen the clinical situation.[14] Therefore, with the result of our study, we hope to alert clinicians and other related disciplines of this under-recognised yet highly prevalent problem in children with ID, especially those in profound grade and studying in special school.

Table 3 Comparison of factors contributing to sleep problem between the "Study group" (total sleep score ≥41) and "Comparison group" (total sleep score <41)

	Study group (n= 56)	Comparison group (n=11)	OR (95% CI)	p-value
Male sex	31/56 (55.4%)	6/11 (54.5%)	1.03 (0.28-3.77)	0.96
Adolescent age group	34/56 (60.7%)	4/11 (36.4%)	2.70 (0.71-10.33)	0.19
Epilepsy	41/56 (73.2%)	4/11 (36.4%)	4.78 (1.22-18.7)	0.02*
On anti-epileptic drugs	37/56 (66.1%)	4/11 (36.4%)	3.41 (0.88-13.11)	0.06
On ≥2 anti-epileptic drugs	19/56 (33.9%)	1/11 (9.1%)	5.14 (0.61-43.16)	0.15
Cerebral palsy	22/56 (39.3%)	6/11 (54.5%)	0.54 (0.15-1.98)	0.35
Bed or wheelchair bound	29/56 (51.8%)	6/11 (54.5%)	0.90 (0.24-3.28)	0.87
Psychiatric problem	4/56 (7.1%)	1/11 (9.1%)	0.77 (0.07-7.62)	0.80
Lives in hostel	29/56 (51.8%)	9/11 (81.8%)	0.24 (0.05-1.21)	0.09
Obesity	7/56 (12.5%)	0/11 (0%)	NS	NS
Presence of medical problem	51/56 (91.1%)	7/11 (63.6%)	5.83 (1.26-27.02)	0.02*

CI = Confidence interval; NS = Not significant; OR = Odd ratio

* p-value ≤0.05

Sleep Disorders and Co-morbidities

A number of co-morbidities and factors are associated with more severe sleep disorders. Those which were previously studied or hypothesised to affect sleep include: age,[14] body weight or obesity,[25] epilepsy,[21,23,24] cerebral palsy or mobility problem,[19] psychiatric diseases (such as autism spectrum disorder),[17,27] visual impairment,[25] active insults (such as acute stroke, head trauma, encephalitis, brain tumour),[32,33] neurodegenerative disorders (such as Parkinson disease, Huntington disease, spinocerebellar atrophy)[34] and neuromuscular disorders (such as muscular dystrophy, congenital myopathy, myotonic dystrophy, myasthenia gravis, hereditary motor sensory neuropathy).[35] For our study, we included the possible factors that affect sleep as listed above in the statistical analysis. There were no subjects suffering from active insults, neuromuscular or neurodegenerative disorders, so these were not included. In such a way, we hoped to identify the risk factors associated with sleep disorders in this group of children.

Our study found that in this group of children with profound ID, there was an association between epilepsy and sleep disorders, and we concluded from the logistic regression model that epilepsy is an independent risk factor for sleep disorders in these children. This association was shown to be unrelated to the use of anti-epileptic drugs. This corroborates results from other studies in children with ID[13,25] as discussed in the introduction. The biological basis of the association between epilepsy and sleep disorders had been extensively investigated in the literature. Epileptic seizure can affect the sleep state, which includes post-ictal hypersomnolence, decreased sleep efficiency, increased wakefulness, sleep stage shifts, arousals and sleep fragmentation.[18] Children with poorly controlled seizure may also have higher percentages of rapid eye movement sleep which is a relatively anti-epileptic state.[17] Clinically, this group of children may be more prone to different types of sleep disorders, including dyssomnias and parasomnias. This will be reflected in different CSHQ sub-scores, which will be elaborated further below (see "sub-score analysis"). In turn, poor sleep quality will also have negative impact in seizure control, as sleep deprivation is well-recognised to lower seizure threshold. Acute sleep deprivation causes an increase of slow-wave sleep, a sleep stage which is highly synchronised. Its slow thalamocortical oscillations may induce onset of seizure.[17] The effect of

Table 4 Logistic regression analysis of risk factors with presence of sleep disorder

Condition	aOR (95% CI)	p-value
Epilepsy	7.99 (1.48-43.25)	0.02*
On anti-epileptic drugs	3.11 (0.19-52.35)	0.43
Cerebral palsy	2.61 (0.43-15.79)	0.29
Wheelchair or bedbound	1.95 (0.28-13.62)	0.50
Psychiatric problem	3.48 (0.24-50.36)	0.36
Lives in hostel	4.64 (0.80-26.86)	0.08
Obesity	0.06 (0.01-3.38)	0.17
Presence of medical problem	2.02 (0.15-26.58)	0.59

CI = Confidence interval; aOR = Adjusted odd ratio

* p-value ≤0.05

Table 5 Comparison of different risk factors on sub-scores

Subscale	Epilepsy			Cerebral palsy			Obesity		
	Present	Absent	p-value	Present	Absent	p-value	Present	Absent	p-value
1	9.20±2.11	8.00±2.33	0.04*	8.64±2.13	8.92±2.33	0.62	8.86±2.26	8.80±2.25	0.95
2	2.00±0.64	1.64±0.71	0.04*	1.93±0.66	1.85±0.71	0.63	2.14±0.69	1.85±0.68	0.29
3	5.31±1.62	4.59±1.68	0.10	5.18±1.68	5.00±1.67	0.66	5.43±2.07	5.03±1.63	0.56
4	5.71±1.47	5.45±1.79	0.54	5.21±1.29	5.92±1.71	0.06	6.00±1.91	5.58±1.54	0.51
5	4.67±1.54	4.09±1.41	0.14	4.43±1.50	4.51±1.54	0.82	5.29±1.49	4.38±1.49	0.14
6	9.56±2.15	9.50±2.11	0.92	8.98±1.99	9.95± 2.14	0.06	9.43± 2.37	9.55±2.11	0.88
7	4.07±1.42	3.41±0.59	0.04*	3.86±1.35	3.85±1.18	0.97	4.71±1.11	3.75±1.23	0.05*
8	12.36±2.81	11.55±2.26	0.24	12.07±2.89	12.10±2.51	0.96	12.57± 2.82	12.03±2.65	0.62

* p-value ≤0.05

Subscale: 1 = Bedtime resistance, 2 = Sleep onset delay, 3 = Sleep duration, 4 = Sleep anxiety, 5 = Night wakings, 6 = Parasomnias, 7 = Sleep-disordered breathing, 8 = Daytime sleepiness

sleep was also evident in some specific epileptic syndromes, such as benign childhood epilepsy with centrotemporal spikes or autosomal-dominant nocturnal frontal lobe epilepsy.[18] Although our study did not demonstrate any cause-effect association between epilepsy and sleep disorders, we believe their relationship is bi-directional. Therefore, it is important to take note that children with ID and epilepsy have a higher chance to develop sleep disorders than those without epilepsy, these children should deserve more attention concerning their sleep. Management should be targeted on both the sleep disorders and epilepsy per se.

Our study failed to identify cerebral palsy as an independent risk factor for sleep disorder in children with ID, in contrast to some previous studies.[19,25] This difference may be attributed to the following: Firstly, our sample consisted of a heterogeneous group of cerebral palsy, from those who could walk without assistance, to those who were bedbound. Those studies which demonstrated the association mainly focused on children with severe locomotor disability. Secondly, it may be due to different case definitions. Some studies included children with neurodegenerative disorder or those with progressive clinical course, which might not be classified as cerebral palsy in strict definition. Thirdly, incontinence care during night-time is also a cause for sleep fragmentation. This practice may be different among institutions or the boarding status of the subject.

We also failed to identify the association between visual impairment and psychiatric disease with sleep disorders. This may be due to low prevalence rates of these disorders in our sample population. In particular for visual impairment, some studies use "blindness", defined as no behavioural responses to visual stimuli, instead. In our sample however, there was no subject fulfilling the criteria of "blindness". It was known that circadian rhythm is heavily influenced by environmental light to the eyes, so this may explain the difference in outcome between different case definitions.

Sub-score Analysis

CSHQ has eight sub-scores, each represents a different type of sleep disorder, including dyssomnias and parasomnias. The analysis of these sub-scores can help us delineate the contributing factors to the underlying whole picture of sleep disorder.

(1) Epilepsy and sleep-disordered breathing. Our result corroborates other studies, showing higher prevalence of sleep-disordered breathing in those with epilepsy than their counterparts.[19,23] Children with profound ID and epilepsy have been shown to have a higher risk of having sleep-disordered breathing associated with hypoxaemia at night.[36] The underlying sleep architecture has been described. The presence of sleep fragmentation, higher percentage of slow-wave sleep and higher total cyclic alternating pattern rate have been reported. It was hypothesised that increased slow-wave activity could be caused by the influence from epileptic discharges, with relation to the thalamo-cortical activity.[26] The use of anti-epileptic drugs can also potentially worsen sleep-disordered breathing, as many of these drugs act on the central nervous system and can suppress breathing during sleep. In turn, sleep-disordered breathing can also worsen the seizure control, as it causes frequent arousal and nocturnal hypoxia, which can potentially trigger a seizure attack. Frequent desaturations causing hypoxaemia is likely to lower the seizure threshold in children with epilepsy. It was shown that treatment of sleep-disordered breathing (such as by adenotonsillectomy) could reduce the seizure frequency in some of the children with profound ID, and it was hypothesised that an improvement in the oxygen saturation could account for the seizure reduction.[20,26]

(2) Epilepsy and bedtime resistance/sleep onset delay. Our result showed that those with co-morbid epilepsy had more problem of bedtime resistance and sleep onset delay, which corroborates with a study showing children with epilepsy had these two CSHQ sub-scores higher than the control group.[21] The disturbance of the circadian rhythm by the underlying epileptic disorder is seen to affect the sleep architecture. This may form a basis for the management strategy of this group of children, by using both medication and non-pharmacological means.

(3) Obesity and sleep-disordered breathing. The association between body weight and sleep-disordered breathing has long been described in the literature. The proposed mechanisms include: narrowed airway (including fat deposits around the upper airway structure and increased pharyngeal collapsibility); altered respiratory mechanics (including decreased chest wall compliance and diaphragm movement); possible blunted hypercapnic ventilatory response causing abnormal ventilatory drive.[37] These mechanisms are more pronounced in children with ID. There is also growing evidence that sleep-disordered breathing can worsen obesity, partly because of excessive daytime sleepiness causing inactivity. It also increases the risk of cardiovascular disease, hypertension, and metabolic abnormalities.[38] Children with profound ID are more prone

to these complications, which should be actively looked for in this population. Sleep behaviour was shown to be associated with obesity, physical activity and sedentary lifestyle in a group of adolescents with ID.[39] These have to be taken into account when managing sleep disorders in this group of children.

Limitation of This Study and Future Direction

The relatively small sample size of this study renders the analysis of subgroup difficult. We also did not include a control group with normal population as comparison. We propose that in future studies this can be included, and the study population can be expanded, such as to include children with different levels of ID.

Conclusion

Sleep disorders are common among children with profound ID, and epilepsy was significantly associated with sleep disorder in this group of children. Correct identification of the sleep disorders and provision of different sleep interventions should be fundamental in the care of these children.

Acknowledgement

We thank MOE - Shanghai Key Laboratory of Children's Environmental Health, Xinhua Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, P. R. China for providing the Chinese version of CSHQ and the scoring system.

We would also like to show our gratitude to the staff and parents of Haven of Hope Sunnyside School for their help and support.

Conflict of Interest

All authors declare no conflict of interest.

References

1. Diagnostic and statistical manual of mental disorders (5th ed.). American Psychiatric Association, Washington, DC, 2013.
2. May ME, Kennedy CH. Health and Problem Behavior Among People With Intellectual Disabilities. Behav Anal Pract 2010;3: 4-12.
3. Childhood Developmental Problems Series. Child Assessment Service, Department of Health Hong Kong Special Administrative Region, 2009.
4. The ICD-10 classification of mental and behavioural disorders: Clinical descriptions and diagnostic guidelines. World Health Organization, 1992.
5. Hong Kong Monthly Digest of Statistics. Census and Statistics Department, Hong Kong Special Administrative Region. Persons with Disabilities and Chronic Diseases in Hong Kong, 2015 January.
6. Student Enrolment Statistics. Education Bureau, Hong Kong Special Administrative Region, 2015/16.
7. Quine L. Sleep problems in primary school children: comparison between mainstream and special school children. Child Care Health Dev 2001;27:201-21.
8. Stores G. Children's sleep disorders: modern approaches, developmental effects, and children at special risk. Dev Med Child Neurol 1999;41:568-73.
9. Sateia MJ. International classification of sleep disorders-third edition: highlights and modifications. Chest 2014;146:1387-94.
10. Wiggs L. Sleep problems in children with developmental disorders. J R Soc Med 2001;94:177-9.
11. Richdale A, Francis A, Gavidia-Payne S, Cotton S. Stress, behaviour and sleep problems in children with an intellectual disability. Journal of Intellectual & Developmental Disability 2000; 25:147-61.
12. Stores G, Wiggs L. Sleep Disturbance in Children and Adolescents with Disorders of Development: Its Significance and Management. Cambridge University Press; 2001.
13. Didden R, Sigafoos J. A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. Res Dev Disabil 2001;22:255-72.
14. Bartlett LB, Rooney V, Spedding S. Nocturnal difficulties in a population of mentally handicapped children. British Journal of Mental Subnormality 1985;31:54-9.
15. Quine L. Sleep problems in children with mental handicap. J Ment Defic Res 1991;35:269-90.
16. Didden R, Korzilius H, van Aperlo B, van Overloop C, de Vries M. Sleep problems and daytime problem behaviours in children with intellectual disability. J Intellect Disabil Res 2002;46:537-47.
17. Malow BA. Sleep disorders, epilepsy, and autism. Ment Retard Dev Disabil Res Rev 2004;10:122-5.
18. Kataria L, Vaughn BV. Sleep and Epilepsy. Sleep Med Clin 2016; 11:25-38.
19. Ming X, Pak J, Mulvey MA, et al. Sleep complaints in cerebral palsy and/or epilepsy: A pediatric sleep questionnaire study. J Pediatr Neurol 2014;12:127-35.
20. Kotagal S, Kothare SV. Sleep in Childhood Neurological Disorders. Demos Medical Publishing, LLC, 2001.
21. Larson AM, Ryther RC, Jennesson M, et al. Impact of pediatric epilepsy on sleep patterns and behaviors in children and parents. Epilepsia 2012;53:1162-9.
22. Boyle A, Melville CA, Morrison J, et al. A cohort study of the prevalence of sleep problems in adults with intellectual disabilities. J Sleep Res 2010;19:42-53.
23. Maganti R, Hausman N, Koehn M, Sandok E, Glurich I, Mukesh BN. Excessive daytime sleepiness and sleep complaints among children with epilepsy. Epilepsy Behav 2006;8:272-7.
24. Becker DA, Fennell EB, Carney PR. Daytime behavior and sleep disturbance in childhood epilepsy. Epilepsy Behav 2004; 5:708-15.

25. Lindblom N, Heiskala H, Kaski M, et al. Neurological impairments and sleep-wake behaviour among the mentally retarded. J Sleep Res 2001;10:309-18.
26. Miano S, Bruni O, Aricò D, Elia M, Ferri R. Polysomnographic assessment of sleep disturbances in children with developmental disabilities and seizures. Neurol Sci 2010;31:575-83.
27. Rzepecka H, McKenzie K, McClure I, Murphy S. Sleep, anxiety and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. Res Dev Disabil 2011;32:2758-66.
28. Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. Sleep 2000;15;23:1043-51.
29. Stuttard L, Beresford B, Clarke S, Beecham J, Curtis J. A preliminary investigation into the effectiveness of a group-delivered sleep management intervention for parents of children with intellectual disabilities. J Intellect Disabil 2015;19:342-55.
30. Li SH, Jin XM, Shen XM, et al. Development and psychometric properties of the Chinese version of Children's Sleep Habits Questionnaire. Chin J Pediatr 2007;45:176-80.
31. Zucconi M, Bruni O. Sleep disorders in children with neurologic diseases. Semin Pediatr Neurol 2001;8:258-75.
32. Dyken ME, Afifi AK, Lin-Dyken DC. Sleep-related problems in neurologic diseases. Chest 2012;141:528-44.
33. Baumann CR. Sleep and Traumatic Brain Injury. Sleep Med Clin 2016;11:19-23.
34. Iranzo A. Sleep in Neurodegenerative Diseases. Sleep Med Clin 2016;11:1-18.
35. Fermin AM, Afzal U, Culebras A. Sleep in Neuromuscular Diseases. Sleep Med Clin 2016;11:53-64.
36. Koh S, Ward SL, Lin M, Chen LS. Sleep apnea treatment improves seizure control in children with neurodevelopmental disorders. Pediatr Neurol 2000;22:36-9.
37. Trang H. Sleep-Disordered Breathing In Obese Children. Mecanisms, Diagnosis and Management. In M.L. Frelut (Ed.), The ECOG's eBook on Child and Adolescent Obesity, 2015. Retrieved from ebook.ecog-obesity.eu.
38. Leinum CJ, Dopp JM, Morgan BJ. Sleep-disordered breathing and obesity: pathophysiology, complications, and treatment. Nutr Clin Pract 2009;24:675-87.
39. Vanhelst J, Bui-Xuan G, Fardy PS, Mikulovic J. Relationship between sleep habits, anthropometric characteristics and lifestyle habits in adolescents with intellectual disabilities. Res Dev Disabil 2013;34:2614-20.