

Study of the Sleep Disturbances in Children with Autism Spectrum Disorder

IMRAN MUSHTAQ¹

Institute of Child Health
Department of Pediatrics
Sir Ganga Ram Hospital, New Delhi
India

PRAVEEN SUMAN

Institute of Child Health
Department of Pediatrics
Sir Ganga Ram Hospital, New Delhi
India

Abstract:

Sleep disturbances often have a significant effect on the clinical manifestations and symptom severity, as well as on the management of neurobehavioral disorders e.g; Autism in children. Children undergoing evaluation for Autism Spectrum Disorder should be routinely screened for sleep disturbances as they are more common in children with autism and may have an impact on the management of these children. This is the first study of sleep disturbances in Autistic Spectrum Disorder on Indian Population. In this study we screened and diagnosed autism in children and studied the subjective measures of sleep in children with Autism aged 3-12 years as compared to healthy controls of the same age group and studied the relation of sleep disturbances and Autism Spectrum Disorder. Diagnosis of Autism was done using DSM IV criteria and severity of autism was assessed by Childhood Autism Rating Scale (CARS). Subjective assessment of sleep was done using Child's Sleep Habits Questionnaire (CSHQ). Children with Autism had significantly higher score (more disturbed sleep) on 5 out of 8 sleep subscales of CSHQ (Parent measure) viz: Bed Time

¹ Corresponding author: imnoorani@gmail.com

Resistance, Sleep Onset Delay, Sleep duration, Sleep Anxiety and Parasomnias than did the controls. Total CSHQ score was significantly higher in children with Autism as compared to normal controls indicative of more sleep problems in autistic children. There was no correlation between sleep disturbances and severity of Autism.

Key words: Autism, Child Development, Sleep Disorders, Developmental Disorders, Autism Spectrum Disorder, Neuropsychology.

Introduction

Autism, first described by American psychiatrist Leo Kanner in 1943 is a disorder of neural development characterized by impaired social interaction and communication, and by restricted and repetitive behaviour. These signs all begin before a child is three years old. According to American Psychiatric Association's Diagnostic & Statistical Manual Of Mental Disorders, Fourth Edition (DSM-1V) Autism spectrum disorders (ASDs) are a group of neurodevelopment disabilities defined by developmental deficits in 3 domains: communication; social and emotional reciprocity; and stereotyped interests, behaviour, and activities.¹ ASDs affect individuals worldwide with a prevalence rate of 60 to 70 per 10,000 as of recent studies.^{2,3} The precise neurobiological causes of autism have yet to be elucidated, but genetic factors play an important role.^{4,5} In about one-third of the children affected, autism becomes apparent after a period of apparently normal or only relatively delayed development, with a loss of previously acquired abilities.^{6,7} The main clinical feature of this phenomenon, which has been defined as "autistic regression," is the loss of spoken language, but social interests and other communicative skills are also generally affected. The Autism Spectrum Disorder (ASD) is one of the most devastating neurobiological disorders of prenatal and postnatal brain development.⁸ The triad of

difficulties or core deficits of ASD include significant impairments in social interaction, limited communication, and a restricted repertoire of behaviours, interests, and activities.⁹

Sleep problems may be intrinsic to autism, such as obstructive sleep apnea syndrome (OSAS), or may be because of extrinsic or environmental factors, such as behavioural insomnia of childhood (sleep onset association type and limit setting type) and insufficient sleep, may manifest primarily with daytime sleepiness and neurobehavioral symptoms. Sleep disturbances often have a significant effect on the clinical manifestations and symptom severity, as well as on the management, of neurobehavioral disorders in children and adolescents. Settling Problems or prolonged sleep latency ,which manifests clinically as difficulty falling asleep and / or bed time resistance and restless or fragmented sleep problems are problematic for children with autism and may result in significantly shortened sleep duration and insufficient sleep as well as decreased total sleep efficiency(time asleep/time in bed).In addition , there is clear evidence from both experimental laboratory based studies and clinical observations that insufficient and poor quality sleep result in daytime sleepiness and behavioural dysregulation and affect neuro-cognitive functions in children.

The goals of our study were: 1) To screen and diagnose children for Autistic Spectrum Disorder. 2) To study the subjective measures of sleep in children with Autism as compared to healthy age matched controls of 3-12 year age group. 3) To correlate the severity of autism and sleep disorders. To accomplish these goals, we characterized sleep in children with ASD using multiple measures, including parental report, a validated sleep questionnaire, and parent reports. We screened the children for Autistic Spectrum Disorder using DSM IV criteria and Childhood Autism Rating Scale. Subjective assessment of sleep was done using Child's Sleep Habits Questionnaire (CSHQ).The parents were counselled for better

sleep hygiene for the children.

Methods:

Participants:

1. Subject Recruitment and Selection:

A sample size of 25 children and a control of 25 was used for the study. This study screened and diagnosed Autism in children with development delays in the age group of 3-12 years and evaluated sleep disorders in children diagnosed with ASD who receive care at the Institute Of Child Health, Department Of Pediatrics, Sir Ganga Ram Hospital New Delhi India. Children, aged 3 to 12 years, were randomly selected from the OPDs of the developmental clinic. Selecting a random sample enabled us to obtain an estimate of the prevalence of sleep disturbances. During the research parent permission was obtained. Children with Autism randomly selected from the clinic were matched by age and sex to subjects from the control group of children.

2. Inclusion Criteria:

Children enrolled as cases and control subjects in the study met the following criteria: age between 3 and 12 years, stable medical and behaviour conditions, and no change in medication related to sleep or health status in the past 3 months. Autism diagnosis was made by a developmental pediatrician along with a clinical psychologist and confirmed by the DSM-IV-TR checklist and Childhood Autism Rating Scale (CARS).

3. Exclusion Criteria:

Exclusion criteria included parents or guardians who were unable or unwilling to provide consent, children with significant hearing or vision loss, and children with a psychiatric disorder established by DSM-IV-TR criteria. The comorbid psychiatric disorders were identified during chart review by investigator. All children in the study were evaluated

for their diagnosis by a developmental pediatrician and a psychologist. Children with co-morbid psychiatric disorders were excluded from the study. In addition, children with a complex neurologic disorder (cerebral palsy, phenylketonuria, tuberous sclerosis, neurofibromatosis, unstable seizure disorder [a seizure within 6 months], Rett disorder) and children with an unstable medical condition (e.g. asthma, diabetes, cystic fibrosis, cardiac disease) were excluded.

Tools:

Sleep Related Measures

Children's Sleep Habits Questionnaire:

Parents completed the Children's Sleep Habits Questionnaire (CSHQ). The CSHQ has been used to examine sleep behavior in children with a variety of conditions, including ASD.^{3,5} Domains on the CSHQ include insomnia-related dimensions such as bedtime resistance, sleep anxiety, sleep onset delay, sleep duration, and night wakings, as well as daytime sleepiness, sleep disordered breathing, and parasomnias. Based on the literature of parental sleep concerns in ASD (see introduction for references), we hypothesized a clinically relevant difference of 40% in prevalence of sleep disorder between cases (Autism) and control group. We needed at least 25 observations per group to achieve 90% power at 5% level of significance as per the formula discussed in statistical analysis. DSM IV checklist for autism was used for screening. The Children were then assessed on Gesell's Development Schedule for delays in development.

Childhood Autism Rating Scale:

The purpose of the Childhood Autism Rating Scale is to identify the presence of autism in children. Fifteen domains are rated on a 7-point Likert scale (assigned values range from 1 to 4 – 1, 1.5, 2, 2.5, 3, 3.5, 4): Relating to people; Imitative behaviour; Emotional response; Body use; Object use; Adaptation to

change; Visual response; Listening response; Perceptive response; Fear and anxiety; Verbal communication; Nonverbal communication; Activity level; Level and consistency of intellectual relations; and General impressions. Ratings from within normal limits to severely abnormal are based on observation, parent interview, and other records. The Total Score, generated from the 15 domains, provides a rating in one of three categories – nonautistic, mild to moderately autistic, or severely autistic. The frequency of mildly-moderate autistic subjects was 76% and for the severe autistic subjects the frequency was 24 % (Table 3) with 15 category rating scores. The total score of 30-36 was mildly-moderately autistic and more than 36 was severe autism. Out of 25 cases 19 had mildly-moderate autism & 6 were severely autistic.

Statistical Analysis:

(A) Sample size calculation:

For our sample size calculation, we defined a clinically relevant difference of 40% in prevalence of sleep disorder between cases (Autism) and control group. We needed at least 25 observations per group to achieve 90% power at 5% level of significance. The formula for calculated sample size is given below:

$$n = \frac{[z_{1-\alpha/2} \cdot \sqrt{2P(1-P)} + z_{1-\beta} \cdot \sqrt{P_1(1-P_1) + P_2(1-P_2)}]^2}{(P_1 - P_2)^2}$$

Where

P1 = Anticipated proportion of sleep disorder in Autism.

P2 = Anticipated proportion of sleep disorder in Controls.

$$P = (P1 + P2) / 2$$

(B) Statistical method:

Continuous variables were presented as mean±SD or medians if they were unevenly distributed. Categorical variables were expressed as frequencies (%). Differences between groups were

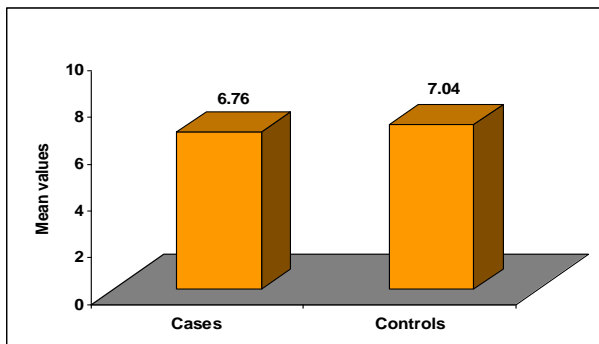
assessed with Chi-square or fisher's exact test for categorical variables. Unpaired t tests were used for comparison of continuous variables between the two groups. For non parametric data, Mann Whitney U test was used. $P < 0.05$ was considered statistically significant. Statistical analysis was performed with SPSS version 17.0 software.

Results:

Table 1. Comparison of Age between Cases and Control Group; Study Group: 25 cases & 25 controls were enrolled in the study.

	Cases (n=25)		Control (n=25)		P value
	Mean \pm SD	Min - Max	Mean \pm SD	Min - Max	
Age	6.76 \pm 2.13	3 - 10	7.04 \pm 2.01	3 - 12	0.635

	Cases	Controls
Age	6.76	7.04

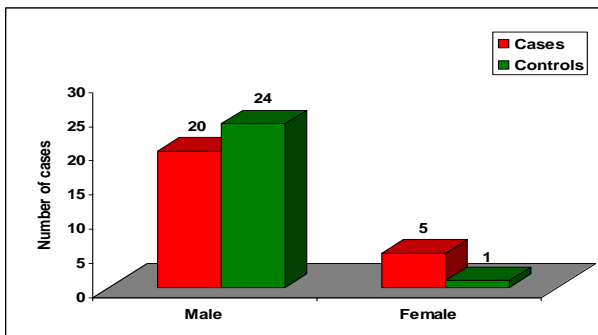


The mean age of cases with autism was 6.76 years and for controls it was 7.04 years. There was no significant difference of age between Autism and Control group (Table 1).

Sex	Cases		Control		P value
	Frequency	%	Frequency	%	
Male	20	80%	24	96%	0.189
Female	5	20%	1	4%	
Total	25	100%	25	100%	

Table: 2 Comparison of sex between cases and control groups.

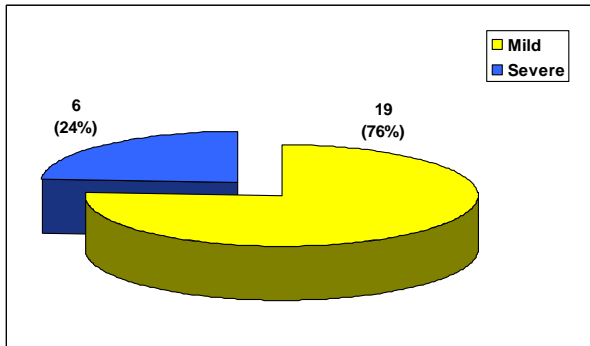
	Cases	Controls
Male	20	24
Female	5	1



The frequency of males and females in the cases was 80% and 20% respectively. For control male and female frequency is 96% and 4%. There was no significant difference in sex between Autism and Control group (Table 2).

Autism	Cases	
	Frequency	%
Mild	19	76%
Severe	6	24%
Total	25	100%

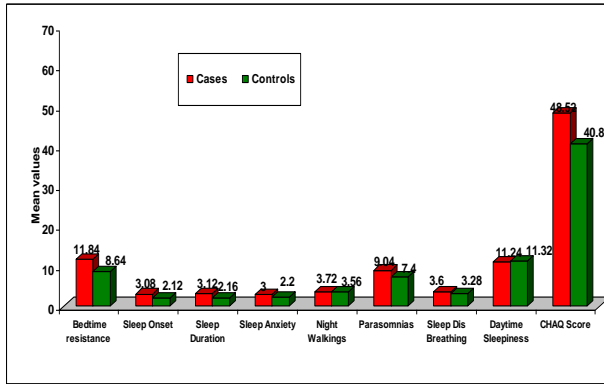
Table 3. Distribution of cases on the basis of severity of autism



	Cases (n=25)		Control (n=25)		P value
	Mean \pm SD	Min - Max	Mean \pm SD	Min - Max	
Bedtime resistance	11.84 \pm 2.15	7 - 16	8.64 \pm 1.35	7 - 13	<0.001
Sleep Onset	3.08 \pm 1.35	2 - 6	2.12 \pm 0.43	2 - 4	<0.001
Sleep Duration	3.12 \pm 1.33	2 - 6	2.16 \pm 1.83	2 - 3	<0.001
Sleep Anxiety	3.0 \pm 1.35	2 - 6	2.20 \pm 0.41	2 - 3	<0.001
Night Waking	3.72 \pm 1.10	3 - 7	3.56 \pm 0.58	3 - 5	0.525
Parasomnias	9.04 \pm 1.84	7 - 13	7.40 \pm 0.50	7 - 8	<0.001
Sleep Disordered Breathing	3.60 \pm 1.44	3 - 8	3.28 \pm 0.46	3 - 4	0.300
Daytime Sleepiness	11.24 \pm 2.63	7 - 19	11.32 \pm 1.28	10 - 13	0.892
CSHQ Score	48.52 \pm 6.44	40 - 66	40.84 \pm 2.32	37 - 46	<0.001

Table 4. Children's Sleep Habits Questionnaire: Comparison of Total and individual Sleep Variable Scores for AUTISM & Control Groups

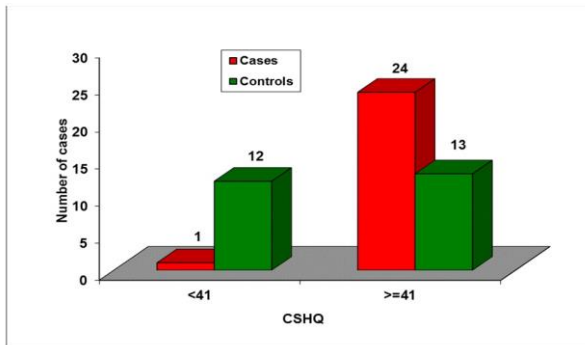
The results were significant on five out of eight scales of CSHQ. The other three results viz; Night waking, Sleep disordered breathing and Daytime sleepiness didn't show significant difference than the controls.



Parents of all children were subjected to Child’s Sleep Habit Questionnaire (CSHQ) {annexure II}. Total score of ≥ 41 on CSHQ was taken as abnormal. Out of 25 cases, all 25 cases (100%) had abnormal score (≥ 41) on CSHQ. The p value is 0.001 which is statistically significant. The Autistic group had a significantly higher score on 5 out of 8 subscales viz: Bedtime resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety and Parasomnias. Thus the difference of sleep disorders in cases and control is significant and the children diagnosed with autistic spectrum disorder have more sleep problems on 5 out of 8 domains of CSHQ.

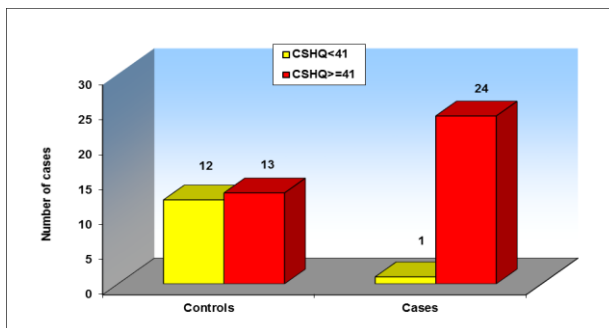
	Cases	Controls
<41	1	12
≥ 41	24	13

Both cases and control were subjected to CSHQ (Childhood Sleep Habit Questionnaire). The scores calculated were indicative of high CSHQ score for cases diagnosed with autism as compared to controls. Out of 25 cases, 24 (96%) were having a score of ≥ 41 which is the abnormal score. Only 1 case (4%) was having score less than 41. In comparison, out of 25 controls, 13 subjects (52%) were having a score of ≥ 41 and 12 controls (48%) were having score <41. Thus, the frequency of sleep problems is higher in cases (autistic children) than that of the controls.



Autism	Controls		Cases		P value
	Frequency	%	Frequency	%	
CHSQ<41	12	48%	1	4%	<0.001
CHSQ>=41	13	52%	24	96%	
Total	25	100%	25	100%	

Comparison of CSHQ Scores and CARS Scores between Control and Cases.



There was significant difference between the CSHQ scores of cases and controls. Our hypothesis of clinically relevant difference of 40% in prevalence of sleep disorder between cases (Autism) and control group is accepted. The prevalence of sleep disorders is more in the children with Autistic Spectrum Disorder than normal controls.

CHAQ	AUTISM				P value
	Mild		Severe		
	Frequency	%	Frequency	%	
<41	0	0%	0	0%	<0.001
>=41	19	100%	6	100%	
Total	19	100%	6	100%	

Comparison of CSHQ Scores and Severity of Autism in Cases

There was significant difference in the sleep problems of children with mild asd as compared to severe asd.

Discussion:

This was a prospective study conducted over a period of 6 months in Child Development Centre, Sir ganga ram hospital New Delhi. This clinic is staffed with full time, experienced Developmental pediatrician, Child Psychologist, Occupational therapist, Speech therapist and Special educator. Twenty five newly diagnosed cases of ASD were enrolled as part of study. Out of twenty five cases, twenty cases were male and five cases were females. There was no significant difference in the results when age and sex was taken as criterion.

Sleep behaviors and sleep quality in children represent a complex exhibition of biologic, developmental, psychological, environmental, and cultural influences. Therefore, careful assessment of a child's sleep behaviors and sleep quality in the context of their family, home, school, and community is paramount.

Children presented with clinical features of impairment in communication, socialization and restricted activities were assessed using DSM IV criteria. While in our study Children who were diagnosed to have Autism by DSM IV-TR criteria were subjected to Childhood Autism Rating Scale(CARS) to know the severity of Autism (non-autistic <30,mildly-moderate 30-36, severely autistic >36). Ratings from within normal limits to severely abnormal were based on observation, parent interview, and other records. The Total Score, generated from the 15 domains, provided a rating in one of three categories –

non autistic, mild to moderately autistic, or severely autistic.

In other studies as well, it has been found that sleep problems are highly prevalent in children with ASD, with rates ranging from 40% to 80%.¹⁰⁻¹⁶ In fact, a recent study found that sleep problems rank as one of the most common concurrent clinical disorders.¹⁷ These sleep problems seem to be relatively specific to children with ASD compared with both typically developing children and children with intellectual disabilities without autism.¹⁸⁻²⁰ A study recently reported that parents of children with ASD had poor sleep quality and decreased quantity compared with parents of typically developing children.²¹

In this study all children were subjected to child sleep habit questionnaire (CSHQ). In CSHQ eight specific sleep parameters were assessed: 1) bedtime resistance; 2) sleep onset delay; 3) sleep duration; 4) sleep anxiety; 5) night waking; 6) parasomnias; 7) sleep disordered breathing; 8) day time sleepiness. Parents were asked to recall sleep behaviors occurred during a recent week. Items were rated on a 3-point scale: 3= "usually" if the sleep behaviour occurred 5-7 times per week; 2= "sometimes" if the sleep behaviour occurred 2 to 4 times per week; 1= "rarely" for 0 to 1 times per week.

The children with Autism were significantly having more impaired sleep than the controls in 5 out of 8 domains of the subjective sleep measures viz: bed time resistance, sleep onset delay, sleep duration, sleep anxiety, and parasomnias. The p value <.001 was found between the scores of CSHQ for cases and controls which according to our hypothesis was accepted as significant.

There was no significant difference between cases and controls for Sleep disordered breathing. Most of the children with Autism in our study were of average weight that might be the reason as Obesity, which has been demonstrated to be the most significant risk factor for the development of obstructive sleep apnea (OSA) in adults, has also been identified as an

increasingly important risk factor for childhood OSA. There was no significant difference between cases and controls because of age and sex as criteria. There was significant difference between the severity of autism and the CSHQ scores. Several studies have suggested that those children with Autism are at greater risk of experiencing impairments related to sleep and thus these children need interventions regarding sleep hygiene.

In summary, we concluded that the children with Autistic Spectrum disorder have more sleep disturbances as compared to the controls. The difference between the sleep problems in cases and controls was statistically significant. The difference between the cases and controls because of age was not significant. The results of this study was in acceptance with our hypothesis of clinically relevant difference of 40% in prevalence of sleep disorder between cases (Autism) and control group. There was significant difference between the severity of autism and the CSHQ scores. The clinician or a psychologist assessing a child with ASD should carefully screen for sleep disorders and make referrals as indicated. Fortunately, there are a variety of treatments available, including behavioural interventions and pharmacotherapy. Identifying and treating sleep disorders may not only result in improved sleep but also favourably affect daytime behavior and family functioning.

Significance of the Study:

Sleep problems are highly prevalent in children with ASD. Identifying and treating sleep disorders may not only result in improved sleep but also favourably affect daytime behaviour and family functioning. Although children with developmental disorders in general have a high rate of sleep problems, children with autism differ within that group. Sleep disturbances may contribute to stress in families of children with ASD and developmental disabilities. Parents of children

with ASD who report sleep problems in their children also report more frequent daily stress and more intense hassles. There is a correlation between sleep problems in children with developmental disorders and maternal stress and parental sleep disruption. Sleep problems may worsen daytime behaviour in individuals with developmental disabilities and in children with Autism. Behavioural issues such as inattention and hyperactivity may be worsened by the presence of sleep disorders such as sleep disordered breathing. In Autism, short sleep duration has been associated with higher rates of stereotypic behaviour, as well as higher overall autism severity scores and social skills deficits. It is critical to identify and address sleep problems in children with Autism because of the effect on health and quality of life in both the children and their parents. The cornerstone of this project is to know whether there is significant prevalence of sleep disorders in autism as identification of this relation between sleep disorders and autism may result not only in formulating more consolidated sleep procedures, more rapid time to fall asleep, and avoidance of night waking but also favourably affect daytime behaviour and parental stress. Addressing sleep problems in children with ASD may not only improve daytime behaviour but also the parents functioning. Targeting effective treatment strategies is dependent on understanding the underlying causes of sleep problems in children with Autism spectrum disorders, therefore further research is paramount. Sleep Concerns are common in children with autism spectrum disorders (ASD). Sleep disorders underlying the sleeplessness were most commonly behavioural (i.e. do with inappropriate sleep-related behaviours). Like children with other developmental disabilities, children with autism spectrum disorders suffer from sleep problems at a greater rate than typically developing children. It is increasingly recognized that addressing sleep problems may improve daytime functioning and decrease family stress. Sleep problems correlate with family distress and

may have significant effects on daytime functioning and quality of life of children with ASDs. In some cases of autism, there may be an identifiable etiology such as obstructive sleep apnea or gastroesophageal reflux; assessment and treatment are guided by history and physical examination. When there is not an identifiable medical cause, behavioural interventions including sleep-hygiene measures, restriction of daytime sleep, positive bedtime routines, and extinction procedures often are effective. This study helped in early identification of autism in developmentally delayed children, and establish a relationship between autism and their sleep disturbances and hence counsel the parents for better sleep hygiene measures and early intervention programs for children.

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