Sleep Profile in Children with Pervasive Developmental Disorders

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Abstract

Sleep disturbances are regarded as a common clinical feature in autism and other Pervasive Developmental Disorders (PDD) that forms a great source of stress for families. Studies have shown that children with PDD exhibited qualitatively and quantitatively different sleep patterns to non autistic control children. This study aimed at describing sleep patterns in a sample of Egyptian children with pervasive developmental disorders. The study included 15 children with a pervasive developmental disorder according to DSM-IV criteria, randomly chosen from the child psychiatry out patient clinic, at the Institute of Psychiatry, Ain Shams University between the months of January and August, 2004. An age and sex matched control group, formed of 10 healthy age and sex matched children from the information bank of the sleep laboratory were obtained. All cases were clinically assessed and the severity of the PDD was further evaluated by the Childhood Autism Rating Scale (CARS). Subjective sleep assessment was obtained through the Arabic version of Children's Sleep Habits Questionnaire (CSHQ). Sleep was also objectively assessed by a polysomnogram performed at the sleep lab of the institute of psychiatry, Ain shams University. The children were classified as mildly autistic or moderately autistic by the CARS. Epilepsy was reported in 46.7% of the patients. Normal sleep latency was reported in 60% of the patients while 26.7% reported a moderate increase in sleep latency. The children's sleep habits questionnaire (CSHQ) showed that sleep duration was adequate in 93.3% of patients. Sleep related anxiety was seen in 53.3% of patients and night awakening in 40% of patients. Nocturnal enuresis was seen in 20% of patients (3 patients) and increased movements during sleep in 26.7% (4 patients). Sleep bruxism was seen in 20% of patients (3 patients), while sleep disordered breathing occurred in one patient. As compared to age and sex matched control, the polysomnogram has shown a significant decrease in sleep efficiency, prolongation of stage 1 and 2 NREM sleep and shortened REM sleep in patients with PDD. The arousal index and number of awakenings were significantly higher in children with PDD than in the control group. The Periodic Leg Movements during Sleep index was also significantly higher in the patient than the control group. The results of our study confirm the presence of sleep disturbances in children with PDD in the form of decreased sleep efficiency and change of sleep

Introduction:

The pervasive developmental disorders (PDD) are a group of disorders in which the main features are delay and deviance in the development of social skills, language and communication, and limited and stereotyped repertoire of behavior and interests. Autism is the prototype of these disorders but the group also includes Asperger syndrome, childhood disintegrative disorder, Rett syndrome and PDD not otherwise specified (Kaplan and Sadock, 1998). The differentiation between these disorders is mainly behavioral with no structural, biochemical or etiological factor identified as specific to any PDD subtype (Willemson-Swinkels and Buitelaar, 2002). The exact causes underlying these disorders are not fully understood but it is believed to

be heterogeneous, with interactions between genetic and environmental factors. A variety of disorders were associated with autism including viral infections, inborn errors of metabolism, structural lesions of the brain, congenital or early neonatal infections, suboptimal obstetric conditions, disturbed absorption from the gastrointestinal tract, altered immunological reactions and endocrinal disturbances. All these factors are believed to act through a final common pathway to affect development of the brain at a critical period resulting in the behavioral syndrome that we call autism (Gilberg and Coleman 2000).

Sleep disturbances are regarded as a common clinical feature in autism and other PDD that forms a great source of stress for families (Herring et al., 1999). Studies have shown that children with PDD exhibited qualitatively and quantitatively different sleep patterns to non autistic control children (Patzold et al., 1998). Villalba and co-workers (2002) classified disorders of sleep in infantile autism into three types:

A) Functional alterations in sleep; with early waking and difficulties in going to sleep being the disorders most frequently seen.

B) Immaturity of sleep: showing a disturbed polysomnographic recording and negative correlations with the level of development.

C) Paroxysmal alterations: with epileptiform discharges being the commonest, without necessarily occurring with seizures.

The exact causes of these sleep disturbances is not fully understood. A variety of factors are believed to play a role including difficulties to regulate sleep and wake cycles according to social clues, altered sensory perception with increased sensitivity to minor environmental changes and increased levels of anxiety (Richdale and Prior, 1995; Patzold et al., 1998). Sleep disturbances may also reflect functional alterations in the neurological structures responsible for regulation of the sleep–wake cycle, and may reflect abnormalities in brain maturation and neurotransmitter systems (Richdale, 1999).

Sleep problems have been correlated with increased personal and family distress and is believed to adversely affect daytime behavior (Patzold et al., 1998) including increased rates of over activity, disruptive behavior, communication difficulties and stereotyped behavior (Patzold et al., 1998; Schreck et al., 2004). Only a few studies have investigated sleep disorders in children with autism and most of these were based on parental reports through sleep diaries or sleep questionnaires. The issue of the objectivity of parental reports was put forth as a potential weakness in various studies. The difficulty inherent in studying autistic children who are intolerant to changes in routine or environment is a likely reason for the paucity of nocturnal polysomnographic studies, even in symptomatic children with an obvious sleep disturbance (Thermulai et al., 2002).

This study aimed at describing sleep patterns in a sample of Egyptian children with pervasive developmental disorders. Children were assessed by means of a clinical history and Childhood Autism Rating Scale (CARS) to diagnose their pervasive developmental disorder and detect its severity. Children Sleep Habits Questionnaire was used to assess sleep subjectively.

Subjects and methods

Study sample:

This study was conducted on 15 Egyptian children, randomly chosen from those attending the child psychiatry out patient clinic, at the Institute of Psychiatry, Ain Shams University between the months of January and August, 2004. Children were diagnosed through clinical history taking according to DSM-IV criteria to have a pervasive developmental disorder. Children below the age of 2 and above the age of 12 were excluded. All children meeting the diagnostic criteria whose parents agreed to participate in the study were included.

A control group, formed of 10 healthy age and sex matched children from the information bank of the sleep laboratory were obtained. The control group was matched to the patients' sex and age.

Evaluation:

A detailed clinical history was obtained from each child including family history, history of perinatal complications, presence of a developmental delay and epilepsy. The age of onset of the pervasive developmental disorders, the clinical features and any associated behavioral disturbances were noted. History of an associated medical or neurological condition in the patient or his family was also included.

The severity of the PDD was further evaluated by the Childhood Autism Rating Scale (Schopler et al., 1993). Evaluation included both an interview with the parent (usually the mother and occasionally both parents) and observation of the child. CARS is a popular tool for screening autistic children. It was shown to correctly identify up to 98% of autistic subjects and 69% of the possibly autistic as autistic (Schopler et al., 1993). It is brief, convenient and suitable for use for any child above the age of two. As most of our patients were nonverbal, and even in those with some language, verbal communication was not possible due to lack of cooperation, non verbal IQ measurements were obtained by trained psychologists.

Subjective sleep assessment was obtained through the Arabic version of Children's Sleep Habits Questionnaire (CSHQ) (Asaad and Kahla, 2001). This is 33 items questionnaire that scores sleep habits of school children as reported by the parents during the past week on a 3 point response scale (often, sometimes, rarely).

The CSHQ yields both a total score and eight subscale scores, reflecting the key sleep domains that encompass the major medical and behavioral sleep disorders in this age group.

Sleep was also objectively assessed by a polysomnogram performed at the sleep lab of the institute of psychiatry, Ainshams University. The child's mother attended the study to comfort the child and put him/her to sleep. The study was also attended by a technician who assured that the electrodes were properly attached all through the study and readjusted them when needed. All medications, except antiepileptics in some patients were stopped before the study.

Statistics:

The data was collected and analyzed with the aid of the program Statistical Package for Social Sciences (SPSS). Quantitative data were described using range, mean and standard deviation. Comparison between the groups was done using Chi square test with Yate's correction.

Results:

The sample included 5 females and 10 males. Their age ranged from 2 to 11 years with a mean age of 5.3 and a standard deviation (SD) of 2.16. Thirteen were diagnosed as autistic disorder, one as Rett syndrome and one as childhood disintegrative disorder.

Family and Past History:

None of the children in this study showed a family history of autism. Perinatal complications were reported in 7 patients (46.7%) and these included Caesarian section (2 patients), breech presentation (1 patient), history of neonatal ICU admission (2 patients), delayed cry (2 patients), low birth weight (2 patients), neonatal cyanosis (2 patients), and maternal gestational diabetes mellitus in one patient.

The developmental history of these children showed normal developmental milestones till the onset of the behavioral disturbance in six of the patients (40%). In the rest (60%), developmental delay in at least one area of development (motor, mental, social or language milestones) was reported.

The medical history of the children revealed one case of Fragile X syndrome, one case of infantile spasms (West syndrome), and one child with history of ambiguous genitalia and dysmorphic features in the form of hypertolerism and low set ears. The chromosomal count of this child was normal.

Epilepsy and Autism:

Epilepsy was reported in 7 (46.7%) patients in this study. Types of seizures included generalized tonic clonic seizures in one patient, adversive fits in two patients, infantile spasms in one patient, generalized tonic seizures in one patient and absence in one patient. Of these, only 5 had an available EEG. The abnormalities detected included bilateral temporal foci in one patient, frontal focus in one patient, generalized epileptic activity in one patient and no abnormality in two patients. Epilepsy was controlled in all patients with no fits occurring in the last month prior to the study.

Behavioral Disturbances:

In 7 patients (46.7%) the parents reported that the behavioral abnormality dated since birth. Onset was before the age of two in 66% and was before the age of three for all the patients in our study. Hyperactivity was reported in 8 patients (53.3%), aggression in 3 patients (20%), and self injurious behavior in 6 patients (40%) in the form of head banging or hand biting, but more severe forms of self injury were not found. The main clinical features of the cases are outlined in Table 1.

IQ:

The IQ of patients in this study ranged from 24 to 75 with a mean of 46.2 and SD of 14.7. Only one patient had an IQ above 70 while all other patients were mentally subnormal. There was no significant correlation between perinatal complications and severity of intellectual disability or incidence of epilepsy. At the same time there was no significant correlation between incidence of epilepsy and perinatal complications.

Results of CARS:

Childhood Autism Rating Scale (CARS) was used to assess the severity of pervasive developmental disorder in the children. The severity of the condition showed no significant correlation to gender. Ratings for the various CARS subscales (table 2).

Results of CSHQ:

The children's sleep habits questionnaire (CSHQ) showed that sleep duration was adequate in 93.3% of patients. Sleep related anxiety was seen in 53.3% of patients and night awakening in 40% of patients. Nocturnal enuresis was seen in 20% of patients (3 patients) and increased movements during sleep in 26.7% (4 patients). Sleep bruxism was seen in 20% of patients (3 patients), while sleep disordered breathing occurred in one patient (table 3).

Parents reported that no or mild bed time resistance was seen in 60% of patients while bed time resistance was moderate in 26.7% of cases and severe in 13.3% of cases. Sleep latency was mildly affected in 73% of cases and moderately prolonged in 6.,7% of cases. Severely increased sleep latency was seen in 20% of patients. Moderate day time sleepiness was seen in 6.7% of patients while mild or no daytime sleepiness was seen in the rest of patients (tables 4).

There were no significant correlations between sleep anxiety or night awakening and severity of PDD.

Results of Polysomnography:

As compared to age and sex matched control, the polysomnogram has shown a significant decrease in sleep efficiency, prolongation of stage 1 and 2 NREM sleep and shortened REM sleep in patients with PDD. The arousal index and number of awakenings were significantly higher in children with PDD than in the control group. The PLMS index was also significantly higher in the patient than the control group. Three of the patients with a periodic leg movement during sleep index (PLMS I) above one were reported by the parents to be hyperactive during the day while three of the patients with a PLMS index above one were reported by the parents to have increased movement during sleep (table 5, figures 1 and 2).

The results of our study showed a highly significant decrease in sleep efficiency and increase in arousal index in patients with moderate PDD than those with mild PDD. This is shown in *table 6*. However, no correlation was found between IQ and either of these sleep parameters. Also there was no correlation between number of awakening, stage 1, stage 2 or REM percentage with either the IQ and the severity of PDD.

Table 1: The main clinical features of the patients

	Present		Absent	
	No	%	No	%
Epilepsy	7	46.7	8	53.3
Hyperactivity	8	53.3	7	46.7
Perinatal complications	7	46.7	8	53.3
Normal early development	6	40	9	60
Aggression	3	20	12	80
Self injury	6	40	9	60

Table 2: Occurrence of various symptoms in the patients as detected by CARS

	Absent%	Mild %	Moderate%	Severe%
Relating to people	13	46	33.3	6.7
Imitation	20	40	26.7	13.3
Emotional response	20	46.7	33.3	0
Body use	33.3	13.3	46.7	6.7
Object use	0	60	33.3	6.7
Adaptation to change	53.3	40	6.7	0
Visual response	20	46.7	33.3	0
Listening response	20	66.7	13.3	0
Touch, smell and taste	46.7	46.7	6.7	0
Fear or nervousness	26.7	40	26.7	6.7
Verbal communication	0	13.3	33.3	53.3
Nonverbal Communication	6.7	26.7	60	6.7
Level of activity	6.7	33.3	53.3	6.7
Intellectual response	6.7	26.7	53.3	6.7
General impression	0	53.3	40	6.7
Total score	0	60	40	0

This table shows the percentage of patients showing mild, moderate and severe symptoms in each of the items of the childhood autism rating scale (CARS).

Table 3: The occurrence of sleep disturbances in patients as detected by CSHQ

	Present		Absent	
	No	%	No	%
Adequate sleep duration	14	93.3	1	6.7
Sleep anxiety	8	53.3	7	46.7
Night awakening	6	40	9	60
Breathing disorders	1	6.7	14	93.3
Nocturnal enuresis	3	20	9	80
Increased movements	4	26.7	11	66.3
Sleep bruxism	3	20	12	80

Table 4: The severities of some sleep parameters in the patients as detected by CSHQ

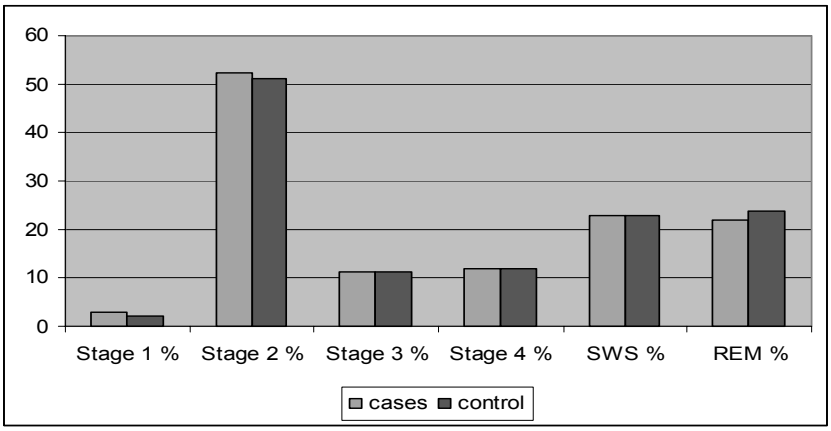
	Mild %	Moderate %	Severe %
Bed time resistance	60	26.7	13.3
Sleep latency	73.3	6.7	20
Day time sleepiness	93.3	6.7	0

Table 5: Polysomnography findings in patient and control groups

Sleep parameter	Cases		Control		t	P	Significance
	Mean	SD	Mean	SD			
Sleep efficiency. %	85.83	4.93	92.66	2.17	4.09	<0.01	HS
Stage 1 %	2.91	0.58	2.03	0.23	4.50	<0.01	HS
Stage 2 %	52.24	1.07	51.20	0.83	2.59	<0.05	S
Stage 3 %	11.36	0.86	11.31	0.34	0.17	> 0.05	NS
Stage 4 %	11.80	0.73	11.96	0.40	0.62	> 0.05	NS
SWS %	22.96	1.23	22.88	0.51	0.19	> 0.05	NS
REM %	21.94	1.15	23.89	1.22	4.01	<0.01	HS
SWSL	27.86	2.97	29.20	1.22	1033	> 0.05	NS
REML	66.46	5.13	67.10	4.01	0.32	> 0.05	NS
REM D	17.48	0.79	17.90	0.44	1.48	> 0.05	NS
Arousal I	0.86	0.39	0.47	0.25	2.73	<0.01	HS
Number of awakenings	1.60	1.12	0.10	.316	4.09	< 0.01	HS
Apnea index	8.66	0.22	0.00	0.00	1.21	> 0.05	NS
Obstructive apnea	6.66	0.17	0.00	0.00	1.16	> 0.05	NS
Mixed apnea	1.33	5.16	0.00	0.00	0.81	> 0.05	NS
Apnea hypoxia index	5.33	0.20	0.00	0.00	0.81	> 0.05	NS
PLMS I	0.92	0.48	0.41	0.24	3.05	<0.05	S

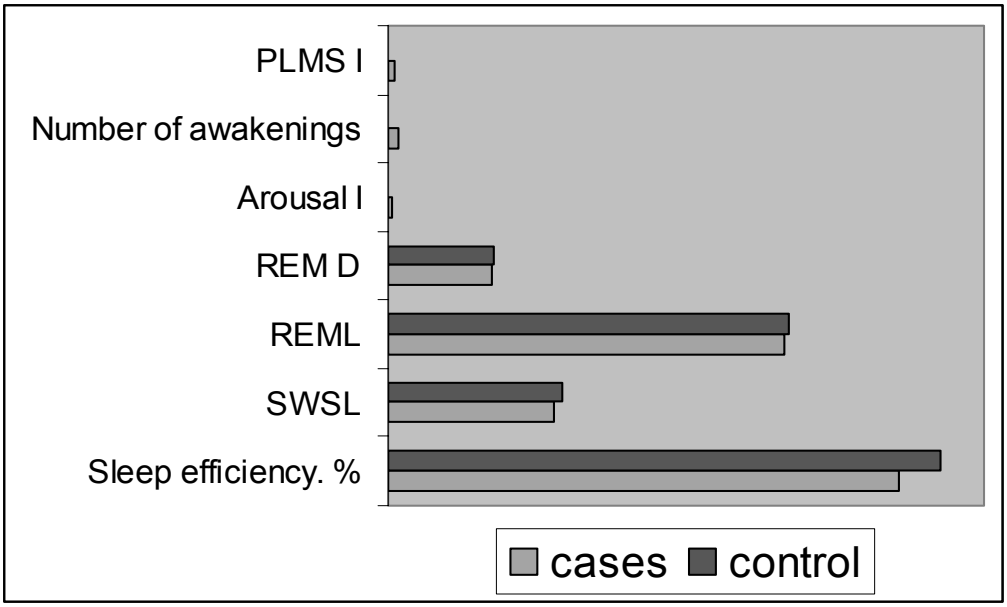
This table compares the various sleep parameters detected by polysomnography in the patient and control groups. REM= rapid eye movement; SWS= slow wave sleep; REML= rapid eye movement latency. SWSL= slow wave sleep latency; REMD= rapid eye movement density ;arousal I = arousal Index. PLMS I= periodic leg movement during sleep index. P>0.05= non significant; P<0.05= significant; P<0.01= highly significant

Figure 1: Comparison between various sleep stages in patients and controls



This figure shows the difference between percentages of different sleep stages in cases and controls. Stages 1 and 2 of NREM sleep are significantly longer while REM sleep is significantly shorter in cases than in control; SWS=slow wave sleep; REM= Rapid eye movements

Figure 2 : Comparison between various polysomnographic parameters in patients and control



This figure shows the difference between different polysomnographic parameters in case and control groups. A significant difference is seen in PLMS index, number of awakening, arousal index and sleep efficiency; PLMS=periodic leg movements during sleep; REM D= Rapid eye movements density, REML= rapid eye movements latency; SWSL= slow wave sleep latency.

Table 6: Polysomnographic findings in patients with mild and moderate autism

	Mild		Moderate		T	P	Significance
	Mean	SD	Mean	SD			
Sleep Efficiency %	87.22	3.78	87.22	5.4	2.00	<0.05	S
Stage 1 %	2.72	0.45	2.72	0.67	1.63	> 0.05	NS
Stage 2 %	52.21	0.56	52.21	1.64	0.15	> 0.05	NS
REM %	22.10	1.21	22.10	1.12	0.61	> 0.05	NS
PLMS Index	0.9	0.54	0.9	0.43	0.18	> 0.05	NS
Arousal index	0.7	0.26	0.7	0.45	2.15	<0.05	S
Number of awakenings	1.33	1.11	1.33	1.09	1.14	> 0.05	NS

This table c*ompares the various sleep parameters detected by polysomnography in patients with mild and moderate autism. A significant difference is seen in sleep efficiency and arousal index. REM= rapid eye movement; PLMS I= periodic leg movement during sleep index. P>0.05= non significant; P<0.05= significant*

Discussion

The pervasive developmental disorders (PDD) are characterized by delay and deviance in the development of social skills, language and communication, and a restricted behavioral repertoire (Sadock and Sadock, 2004). Children with PDD show a number of associated behavioral disturbances including sleep disturbances that are quantitavely and qualitatively different from those exhibited by normal children and those with other psychiatric disorders (Patzold et al., 1998).

Despite the high prevalence of sleep disturbances in children with PDD, only a few polysomnographic studies in autistic children are available in the literature (Tanguay, 1976; Thermulai et al., 2002; Sun et al., 2003) and the number of subjects in each of these studies is small (8 to 17 patients). An even smaller number of studies included polysomnography in Asperger syndrome (Godbout et al., 2000; Tani et al., 2004) and no studies included children with childhood disintegrative disorder or PDD-NOS. Rett syndrome was more extensively studied (Segawa and Nomura, 1990, Espinar-Sierra et al., 1990, Fujino and Hashimoto 1990; Segawa and Nomura, 1992; Marcus et al., 1994; Kohyama et al., 2001).

Our study aimed at describing sleep patterns in a sample of Egyptian children with pervasive developmental disorders. Children were assessed by means of a clinical history and CARS to diagnose their PDD and detect its severity. Children Sleep Habits Questionnaire was used to assess sleep subjectively. The parents are instructed to answer the questionnaire based on the child's sleep habits in the last week. This allows evaluation of the child's sleep habits at home over a relatively long period of time which could not be assessed by the polysomnogram alone. It is also useful for

evaluating events that do not occur every day as nocturnal enuresis and other parasomnias. This was followed by objective sleep assessment by polysomnography. This allowed studying sleep architecture and verifying the data obtained by the questionnaire.

Fifteen children (10 males and 5 females) were chosen randomly from the Child psychiatry out patient clinic of the institute of psychiatry, Ain Shams University Hospital. The male to female ratio in our sample is consistent with various epidemiological studies showing a higher prevalence of autism in males (Fombonne, 1998). The relatively young mean age of the patients (5.3 SD2.16) is well suited to the aim of this study as sleep problems are reported to be more common in younger children (Patzold et al., 1998). Of the children in this study 13 were diagnosed as autistic disorder, one met the criteria for childhood disintegrative disorder and one was diagnosed as Rett syndrome.

Two of the children in our study had a known associated medical condition. One of the children had Fragile X syndrome and the other West syndrome. Another child had dysmorphic features and ambiguous genitalia which are highly suggestive of a chromosomal abnormality. Chromosomal count was normal in this child but a more detailed study for structural chromosomal abnormalities was not available and these can not be excluded. These findings reflect the heterogeneous nature of autism and are to be expected in any clinical sample. Both fragile X and West syndrome were previously associated with sleep abnormalities. For both our patients, different sleep parameters were within one standard deviation from that obtained for the PDD group as a whole, and no specific features could be detected.

Mental sub-normality is a common associated feature in PDD that has been previously associated with disturbances in sleep. Polysomnographic studies showed a reduction of REM sleep percentage, a prolonged latency of the first REM period, a reduction of the number of REM cycles and the presence of undifferentiated sleep in mentally subnormal children. The lack of high functioning autism in our sample, with only one child with an IQ above 70, and the small sample size made it impossible to isolate the effect of mental retardation on the results of our study.

Epilepsy, a common co-morbid condition in patients with PDD, may also affect sleep pattern. In this study, 43.3% of children were reported to have epilepsy. Although chronic epilepsy was previously reported to cause disturbances in sleep architecture (Shouse, 1994), most of the effect was attributed to seizures occurring on the night of the study. There is no generalized agreement considering the influence of seizures taking place prior to the night of the study on the sleep pattern (Lopez Gomez et al., 2004). Most of our patients are well controlled and none of them had any seizure in the month before the study, thus the effect of epilepsy on sleep architecture is expected to be minimal. Five of our patients were on antiepileptic medication on the time of the study including valproate and carbamazepine, and ACTH injections. Carbamazepine was previously shown to decrease REM sleep as well as the frequency and duration of periods of wakefulness while sodium valproate increases deep sleep in children (Nicholoson, 1994). Most available studies on the effects of antiepileptic medications on sleep were carried on epileptic patients

and it is not clear whither the effects of these drugs are due to a primary effect on sleep architecture or to suppression of epileptic activity. All other drugs taken by the patients were stopped at the night of performing polysomnogram. However, the chronic effects of medication or effects of withdrawal of medications on sleep can not be eliminated.

Children in our study were classified as mildly autistic or moderately autistic by the CARS. The lack of severely affected patients is probably due to the small sample size. The most commonly encountered symptom was disturbance in verbal communication (severely affected in 53.3% and moderately affected in 33.3%). This could be explained by the fact that delayed language development is the most common presenting feature of autism (Campbell and Shay, 1995) and the most alarming to the parents.

In previous studies parents reported a variety of sleep disturbances in autistic children, with disorders in initiation and maintenance of sleep being the most common. These manifested as extreme sleep latencies, shortened sleep times and frequent awakening (Thermulai et al., 2002). However, most of the patients (60%) in our study were reported to have within normal sleep latencies by their parents and 26.7% reported a moderate increase in sleep latency. The parents also reported that 93.3% had adequate sleep hours although 40% of the children woke up at least once during the night. It is not clear whither the differences between the results of our study and previous reports reflect a true difference in the pattern of symptoms, a cultural difference in sleep habits or a difference in parental report and reaction to their children's behavior.

Sleep related anxiety was reported in 53% of patients in our study and this showed no correlation with severity of autism. Anxiety is a prominent feature in many children with autism and may contribute to sleep problems (Richdale, 1999). The role of anxiety was believed be more in older children and those with a higher IQ (Richdale and Prior, 1995) and in patients with Asperger syndrome (Tani et al., 2004). The result of our study indicates a high level of anxiety even in younger children with low IQ.

Seven (43.3%) of the children in our study were reported to have a paroxysmal event during sleep by the parents. This is consistent with a large study conducted by Yu and Miles on 163 patients with autism in which parasomnias occurred in 77.3% of patients (Yu and Miles, 2002). In our study three cases of sleep bruxism (20%), three cases of nocturnal enuresis (20%), and four cases (26.7%) of increased movements during sleep were reported by the parents. The polysomnogram showed a significant increase in PLMS index in patients with PDD compared to control.

Bruxism, or the intermittent grinding or clenching of teeth during sleep is a common phenomenon. Yu and Miles found bruxism in 24.5% of patients with autism (Yu and Miles, 2002) which is consistent with the results of our study. The exact etiology of bruxism is not known, however, pharmacologic evidence suggests that the central dopaminergic system may be involved in the pathophysiology of sleep bruxism. Recent studies indicate that bruxism may represent a mild manifestation of REM sleep behavior disorder (RBD). This is particularly interesting in the light of the recent detection of RBD, another dopamine dependent disorder, in five

children with autism and insomnia (Thermulai, 2002).

Periodic limb movements are defined as involuntary repetitive movements that occur primarily during stage 1 and 2 sleep. Our study has shown a significantly higher PLMS index in children with PDD than the control group. However, it was not elevated enough to diagnose PLMS in any of these children. Previous studies have also shown an increased incidence of PLMS in children with autism (Thermulai, 2002, Schreck, 2004). PLMS was previously associated with ADHD and Sun et al reported PLMS in a child with autism and comorbid ADHD (Koherman and Carney, 2000; Sun et al., 2003). Of the seven children with an elevated PLMS index in our study, three were reported by the parents to have increased movements during sleep, but no statistically significant association was found between increased daytime activity and PLMS index.

Although these problems appear as separate items, sleep bruxism, PLMS and RBD are all related to disturbed motor control during sleep. This raises the probability of a specific type of sleep related impairments in CNS motor areas for children with autism and mental retardation (Schreck and Mulick, 2000). Other features that may reflect abnormal motor control during sleep were previously reported in children with autism, including increased dispersed rapid eye movements occurring out of bursts of rapid eye movements, an increased amount of muscle twitches as well as presence of rapid eye movements during stages 1 and 2 of NREM sleep (Diomedi et al., 1999).

Bruxism may be related to hyper function of dopamine, while PLMS may be related to hypofunction of dopaminergic neurotransmission (Montplaisir et al., 1994). Abnormalities in dopamine turnover have been detected in patients with PDD (Takahashi et al., 2001). A hyperdopaminergic function of the CNS might explain the hyperactivity and stereotyped behavior in autism and the response to dopamine receptor antagonists as haloperidol (Kaplan and Sadock, 1998). On the other hand, PET studies have demonstrated low medial prefrontal dopaminergic activity in some patients with autism (Herring et al., 1999).

Nocturnal enuresis was found in three (20%) of the children in our study. High frequencies of nocturnal enuresis were previously reported in children with PDD (Yu and Miles, 2002; Sun et al., 2003). The higher frequency of nocturnal enuresis in children with PDD can thus be explained as part of the general delay of development in these children. The findings in our study are consistent with this explanation as the three enuretic children showed delayed developmental milestones.

Our study has shown a highly significant decrease in sleep efficiency and increase in night awakening in patients with PDD than in the control group. This is in agreement with most of the previous studies on autism (Wiggs and Stores, 2004). Our study also showed a highly significant decrease in sleep efficiency and increase in arousal index in patients with moderate autism than those with mild autism, but no correlation with IQ. The correlation between these parameters and the severity of autism may reflect either a true worsening of the sleep efficiency with more severe autism, or an increased sensitivity of more severely autistic children to the changes in environment.

Sleep architecture showed prolonged stage 1 and 2 NREM sleep percentages and

decreased REM sleep percentage as compared to control. Changes in sleep architecture were previously reported in children with autism, but there are some controversies in the results of different studies.[15,16] Sun et al found a decreased REM percentage which is in agreement with the results of our study.[18] Unlike findings of Elia et al and Diomedi et al, there was no difference in REM density between the patient and control group in our study and no correlation between REM % and IQ. The prolongation in stages 1 and 2 seen in our study has not been previously reported (Diomedi et al., 1999; Elia et al., 2000).

The establishment of a mature sleep wake rhythm is a developmental phenomenon and this could account for the greater prevalence of sleep disorders in children with developmental disabilities in general. Significant sleep fragmentations, manifested by frequent awakenings and arousals were detected in children with PDD in our study. Although hyperactivity is a common symptom in autism, occurring in 53.3% of patients in our study, sleep patterns similar to those previously reported in ADHD were not found. This probably reflects the difference in the pathological and biochemical nature of the two disorders.

The results of our study thus confirm the presence of sleep disturbances in children with PDD. The study of sleep in children with pervasive developmental disorders may be rewarding in more than one way. First, it helps the families deal with a disturbing symptom. Second, sleep disturbances may affect daytime achievement. A recent study by Schreck and co-workers (2004) has shown that sleep problems predicted more intense symptoms of autism. In addition sleep problems have been consistently shown to negatively influence learning rate and cognitive performance in typically developing children and adults. Eliminating sleep problems, whatever their cause may aid these children achieve their full potential.

References

Kaplan H; Sadock B; (1998): Synopsis of psychiatry, Williams and Wilkins; Twenty sixth edition.

Willemson-Swinkels, Buitelaar J.K (2002): The autistic spectrum: subgroups, boundaries and treatment. Psychiatric clinics of North America, Dec 25(4): 811-36.

Gilberg C., Coleman M.(2000): The biology of the autistic syndromes. McKeith press;, Third edition.

Herring E., Epstien R., Elroy S., Iancu D.R., Zelnick N.(1999): Sleep patterns in autistic children. Journal of autism and developmental disorders, Apr 29 (2):143-7.

Patzold E, Richdale A. and Tonge A (1998): An investigation into the sleep characteristics of children with autism and Asperger syndrome. Journal of pediatrics and child health, , 34: 528-33.

Abrill Villalba B., Mendez Garcia M., Sens Capdurla O., Validizan Uson J.R. (2002): Sleep in infantile autism. Rev. Neurol.;, April 16: 641-44.

Richdale A.L., Prior M.R.(1995): The sleep wake rhythm in children with autism. European child and Adolescent psychiatry Jul 4(3): 175-86.

Richdale A. (1999): Sleep problems in autism, prevalence, cause and intervention. Developmental medicine and child neurology, 41: 60-66.

Schreck K.A., Mulick J.A., Smith A.F (2004).: Sleep problems as possible predictors of intensified symptoms of autism. Res Dev Disabil. Jan-Feb; 25(1): 57-66.

Thermulai S.S, Shubin R.A, Robinson R. (2002): Rapid eye movement sleep behavior disorder in children with autism. J. Child Neurol. , Mar.17 (3): 173-8.

Schopler E, Reichter D.J., Rochen-Renner (1993): The childhood autism rating scale (CARS), Published by Western psychological Publishers and distributors.

Asaad T. and Kahla O (2001).: Psychometric sleep assessment instruments: An Arabic version for sleep evaluation, Elnahda, El Fagala, Egypt..

Sadock B.J, Sadock V.A. (2004): The pervasive developmental disorders in Kaplan and Sadock's synopsis of psychiatry:Behavioral science and clinical psychiatry, ninth edition , Lipincott Williams and Wilkins,1208-1231.

Tanguay P.E, Ornitz E.M, Fasythe A.B, Ritvo E.R. (1976): REM activity in normal and autistic children during REM sleep. Journal of autism and childhood schizophrenia, Sep (6) 3: 275-88.

Diomedi M., Curatolo P., Scalese A., Placidi F., Correlo F., Gigli G. L (1999).: Sleep abnormalities in mentally retarded autistic subjects, Down syndrome with mental retardation and normal subjects. Brain Dev., Dec., 21(8): 548-53.

Elia M., Forri R., Musumici S.A., Del Garcio S., Bottitta M., Scudenu C., Miano G., Panerai S., Bertrand T., Gruber J.C. (2000): Sleep in subjects with autistic disorder, a neurophysiological and psychological study. Brain Development, Mar22 (2): 88-92.

Validizan Usan JR; Abril Vilalba B; Mendez Garcia M., Sans Capdevila O. (2002): Nocturnal polysomnogram in childhood autism without epilepsy; Review Neurologie;, Jun. 16, 34(12): 1101-5.

Sun Y. I., Ming S.V., Walter A.S. (2003): Polysomnographic analysis of sleep disrupted autism patients. Sleep, Vol 26, Abstract supplement: A 138.

Tani P, Lindberg N, Nieminen-von Wendt T, von Wendt L, Virkkala J, AppelbergB, Porkka-Heiskanen T (2004).: Sleep in young adults with Asperger syndrome. Neuropsychobiology.; 50(2):147-52.

Godbout R., Bergeron C., Limognes E., Stip E., Motlran L.(2000): A laboratory study of sleep in Asperger's syndrome. Neuroreport; Jan .17, 11: 127-30.

Marcus C.L., Carroll J.L., McColley S.A., Loughlin G.M., Curtis S., Pyzik P., Naidu S. (1994): Polysomnographic characteristics of patients with Rett syndrome. J .Pediatr. Aug; 125(2): 218-24.

Segawa M., Nomura Y. (1990): The pathophysiology of the Rett syndrome from the standpoint of polysomnography. Brain Dev.; 12(1): 55-60.

Espinar-Sierra J., Toledano M.A., Franco C., Campos-Castello J., Gonzalez-Hidalgo M., Oliete F., Garcia-Nart M (1990).: Rett's syndrome; a neurophysiological study. Neurophysiol. Clin. ; Apr; 20(1):35-42.

Fujino K., Hashimoto T. (1990): Studies on the Rett syndrome. Part 2. Polysomnographic and neuroendo-crinological studies; No to Hattatsu. Jan; 22(1):16-23.

Kohyama J., Ohinata J., Hasegawa T. (2001): Disturbance of phasic chin muscle activity during rapid-eye-movement sleep.

Brain Dev,Dec. 23 Suppl 1: 104-7.

Segawa M., Nomura Y. (1992): Polysomnography in the Rett syndrome Brain Dev. .May; 14 Suppl: S46-54.

Fombonne E. (1998): Epidemiological surveys in autism in Autism and pervasive developmental disorders, Edited by Vokmar F. R., Cambridge monographs in child and adolescent psychiatry, Cambridge University Press,:33-53.

Shouse M.N. (1994): Epileptic seizures manifestations during sleep. Principles and practice of sleep medicine; edited by Kreuger, Roth and Dement, Second edition, WB Saunders,: 589-595.

Lopez Gomez E., Hoyo Rodrego B., Rodriguez Nieto I (2004): The effects of epileptic seizures on sleep architecture. Rev Neurol. 2004, Jan. 16-31; 38 (2): 176-80.

Nicholoson A.M., Bradley C.M., Pascoe P.A (1994).: Medication effect on sleep and wakefulness. Principles and practice of sleep medicine; edited by Kreuger, Roth and Dement, Second edition, WB Saunders: 589-595.

Campbell M., Shay J (1995): Pervasive developmental disorders in Kaplan and Sadock Comprehensive Textbook of Psychiatry; William and Wilkins, Sixth edition,: 2277-2293.

Yu P., Miles J.H. (2002): Autism; characterization of sleep disorders; Poster session -Research Day, University of Missouri Health Care.

Koherman M.T., Carney P.R. (2000): Sleep related disorders in neurologic disease during childhood. Pediatric neurology, Vol 23 No 2: 107-113.

Schreck K.A., Mulick J.A. (2000): Parental report of sleep problems in children with autism. J.of Autism Dev. Disord., Apr;30(2): 127-35.

Montplaisir J., Godbout R., Pelletier G., Warnes H (1994): Restless leg syndrome and periodic limb movement during sleep. Principles and practice of sleep medicine; edited by Kreuger, Roth and Dement, Second edition, WB Saunders, 589-595

Takahashi K., Tastenson R., Danfors T., Eeg-Olfssen O., Von Knorring A.L., Moulder R., Engler H., Hartvig D., Ingestrim B., Watanabe Y. (2001): Autism Abstracts presented at society for neuroscience, , Nov, Sandiego CA.

Wiggs L., Stores G. (2004): Sleep patterns and sleep disorders in children with autistic spectrum disorders, insights using parental report and actigraphy. Developmental medicine child neurology, Jun 46 (6): 372-8.

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دراسة لحالة النوم في الأطفال المصابين باضطرابات النوم المتجاوبة

أن أضطرابات النوم تعتبر من الخصائص الإكلينيكية الشائعة في مرضى ذاتية التركيز ومرضي اضطرابات النمو المتجاوز الأخرى و تعتبر من أهم الأسباب المقلقة و المزعجة لعائلات هؤلاء المرضي وهناك العديد من الدراسات السابقة تشير إلي أن مرضي اضطرابات النمو المتجاوز المختلفة لهم خصائص وطراز في النوم مختلف اختلاف نوعي و كمي عن الأطفال الغير مصابين وتهدف هذه الدراسة إلى وصف الخصائص المميزة للنوم عند عينة من الأطفال المصريين المصابين بأضطرابات النمو المتجاوز المختلفة. فقد شملت هذه الدراسة خمسة عشر طفلا ؛ مصابين بأضطرابات النمو المتجاوز المختلفة اختيروا من العيادات الخارجية في مركز الطب النفسي بكلية الطب – جامعة عين شمس في الفترة ما بين شهر يناير و شهر أغسطس لسنة ٢٠٠٤ كما شملت هذه الدراسة علي عشرة من الأطفال الأصحاء الغير مصابين بأي مرض و متماثلين مع المرضي في العمر و الجنس. كل الحالات تم فحصهم إكلينيكيا لتحديد نوع و شدة الإصابة بأي من أضطرابات النمو المتجاوز المختلفة وأكثر من ذالك تم تقدير شدة الإصابة بأي من هذه الأضطرابات عن طريق مقياس تصنيف الأطفال المصابون بذاتية التركيز. قيم أيضا طبيعة النوم عند حالات هذه الدراسة عن طريق النسخة العربية للاستطلاع علي سلوك الأطفال أثناء النوم وبطريقة أكثر موضعية عن طريق جهاز مقياس النوم بمركز الطب النفسي بكلية الطب – جامعة عين شمس. وقد أوضحت نتائج هذه الدراسة أن مرضى ذاتية التركيز مقسمين إلي مجموعة مصابة إصابة بسيطة و مجموعة مصابة إصابة متوسطة وان ٤٠,٧ % من المرضي مصابين بالصرع و ٥٠% لديهم زمن كمون نومي طبيعي بينما ٢٥,٧% لديهم ارتفاع متوسط في زمن الكمون ألنومي. أيضا أوضح استطلاع سلوك الأطفال أثناء النوم إلي أن ٩٣,٣% يتمتعون بمدة نوم كافية و ٢٥,٧% لديهم ارتفاع في نسبة الحركة إثناء النوم و ٢٠% لديهم صرير الأسنان أثناء النوم بينما اضطرابات التنفس أثناء النوم حدثت في مريض واحد فقط (٥,٧%). كما أوضحت مقارنة مجموعة الأطفال المرضي بمجموعة الأطفال الطبيعيين بواسطة جهاز مقياس النوم وجود نقص ذو معني أحصائي في كفاية النوم وطول في زمن المرحلة الأولي و الثانية من شق النوم المتميز بعدم وجود حركة سريعة للعين و قصر في زمن شق النوم المتميز بوجود حركة سريعة للعين عند مرضي أضطرابات النمو المتجاوز المختلفة وذالك بمقارنتهم بالأطفال الغير مصابين. وأيضا أوضحت الدراسة أن معدل حركات الساق الدورية إثناء النوم اعلي بشكل إحصائي واضح في مرضي أضطرابات النمو المتجاوز المختلفة عن الأطفال ألأصحاء. وعلي ذالك فقد خلصت الدراسة إلي أن الأطفال المصابين بمرض أضطرابات النمو المتجاوز يعانون من اضطرابات في النوم علي شكل نقص في كفاية النوم و تغيرات واضحة في طبيعة النوم وبمعرفة هذه الأضطرابات وطبيعتها يمكن مساعدة أهل هؤلاء المرضي علي التعامل مع أطفالهم كما أن علاج هذه الأضطرابات في النوم تساعد علي زيادة معدل تعليم هؤلاء الأطفال وزيادة قدراتهم المعرفية.