REFERRALS TO THE SLEEP CLINIC FOR INDIVIDUALS WITH INTELLECTUAL DISABILITY

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INTRODUCTION

Sleep problems are common in children and adults with intellectual disability (ID). Prevalence rates up to 80% are reported in children with ID up to 16 years of age. High rates of sleep problems persist into adulthood[1]. These problems are persistent and often last for years. Parents and professional caregivers may not ask for advice or help, because they hold the belief that sleep problems are part of the disability. Sometimes they are told by professionals that nothing can be done to solve these problems.
On the initiative of the Gouverneur Kremers Center two multidisciplinary outpatient sleep clinics are functioning at this moment. Each clinic is specialized in diagnosis and treatment of sleep problems in children and adults with ID. Before the exploratory interview and the first visit to the sleep clinic the patients' sleeping habits are examined with the Sleep Questionnaire (see Methods), the Children's Sleep Hygiene Scale (CSHS; translated by Van der Heijden et al[2]) and a sleep diary (kept for 14 consecutive days). Also endogenous melatonin levels in saliva were obtained. In this paper we will discuss results of the Sleep Questionnaire.

METHODS

Patients:
All individuals referred to one of our outpatient sleep clinics for individuals with ID in 2005-2006 were included. Individuals were referred mainly by general practitioners, pediatricians and physicians for individuals with ID.
Sleep questionnaire:
Parents or professional caregivers were asked to fill out the Sleep Questionnaire. This questionnaire was adapted from Wiggs et al[3] and Didden et al[4,5]. The questionnaire consisted of five parts. Part one addressed demographic information (e.g., presence of seizure disorders, breathing problems and level of ID). The second part covers current (i.e. last month) behaviors related to settling to sleep, night waking and early morning waking. In part three, parents were asked to fill in at what times their child usually goes to bed and wakes up in the morning. The fourth part assessed the frequency of occurrence of several behaviors related to sleep (e.g., "Grinds teeth in sleep", "Reluctant to go to bed") on a 7-point scale, from "Never" to "Daily". Finally, the last part contains items about parents' impression of

their child's current or past sleep problems, as well as previous treatment of the child's sleep problem and family's sleep.

Definition of a severe sleep problem:

Criteria for the definition of a severe sleep problem were established by Wiggs et al[3] and Didden et al[4,5]. Three types of sleep problems were distinguished. Severe settling problems occurred three or more nights a week, whereby the individual took more than 1 hour to fall asleep and parents were disturbed during this time. Night waking was defined as severe if it occurred three or more nights a week, and if the individual woke up for more than a few minutes and disturbed parents during that time (e.g., co-sleeping, crying). Finally**,** early morning waking was defined as severe if the individual woke up before 5:00 a.m. and stayed awake during three or more nights a week. A severe sleep problem was diagnosed if an individual had at least one of the above three types of sleep problems and these problem lasted for more than 6 months.

RESULTS

In 2005 and 2006 55 individuals were seen in one of our outpatient sleep clinics (30 males, 25 females). The mean age was 9 years and 5 months (range: 2 yrs 2 months to 39 yrs 5 months). Forty eight patients lived at home with their parents, 3 in a group home and 4 in a residential facility. Seven patients were 18 years or older and 1 of them lived at home with parents. During the day 28 children went to a special daycare center, 7 patients went to an adult activity center, 16 children attended special education, 1 child attended preschool special education, 1 adult worked in a sheltered shop and 2 young children stayed at home.

Table 1 shows that 22 of the individuals referred to the sleep clinic (40%) had a genetic syndrome. Thirteen of the referred individuals (24%) were diagnosed with Autism or Autistic Spectrum Disorder.

Table 1. Primary diagnosis intellectual disabilities

Aethiology	*n*
Autism/Autistic spectrum disorder	13
Genetic syndrome	22
Down syndrome	6
Angelman syndrome	3
Smith-Magenis syndrome	3
Rett syndrome	2
Mowat-Wilson syndrome	1
Rhizomelic Chondrodysplasia Punctata (RCDP)	1
Isodicentric chromosome 15 (IDIC 15)	1
Ring chromosome 22	1
22q13 deletion syndrome	1
18q deletion (mosaic)	1
16 micro deletion	1
Translocation 4;16 (mosaic)	1
Other known causes	7
Cerebral palsy	5
Hydrocephalus	3
West syndrome	1
Intellectual disability of unknown cause	11

Level of ID was known for 28 patients, see Table 2 for more information. None of the patients was blind. Seven patients (13%) were wheelchair dependent. Seventeen patients (31%) had epilepsy and four (13%, n = 30) had reflux as far as parents or professional caregivers knew.

Table 2. Level of intellectual disability (n = 28)

Level of ID	*n*
Borderline (IQ > 70)	1
Mild (IQ 55-70)	6
Moderate (IQ 40-55)	8
Severe ((IQ 24-50)	6
Profound (IQ < 25)	7

At referral twenty five patients (45%) already had medication related to sleep problems of whom 20 used melatonin (dosage range: 1.0 to 9.0 mg). Two patients used alimenazine, 1 patient used alprazolam, 1 patient used pipamperone and 1 patient used sodium valproate for their sleep problems. Four patients took a combination of medications related to sleep problems (melatonin and amitriptyline; melatonin and alimenazine; melatonin and temazepam if necessary; alimenazine and temazepam if necessary). One patient (aged 5 yrs 6 months) used 5 mg melatonin at referral, but parent mentioned that she had used 15 mg in the past (when she was a 3-year-old). Eleven patients (20%) already received medication related to epilepsy of whom 3 also took medication related to sleep problems (i.e. melatonin, n = 2; alimenazine, n = 1).

Table 3. Type of advice or treatment received for sleep problems before referral to the sleep clinic (n = 37)

Type of help	*n*
Education/General information	17
Medical-operation[a]	2
Medical-medication	28
melatonin	20
alimenazine	7
promethazine	5
temazepam	3
diazepam	2
pipamperone	2
lorazepam	1
oxazepam	1
alprazolam	1
sodium valporate	1
amitriptyline	1
omeprazol	1
name of sleep medication not specified	2
Medical-other[b]	2
Psychological/behavioral treatment	8
Other	15
homeopathic	10
Sensory Integration Therapy	2
swaddle blanket	1
Bach flower remedy	1

[a] Drainage tube (grommet) placed/adenoidectomy; [b] Hospitalization/dental checkup.

Parents or professional caregivers of 37 patients (69%) had received advice or treatment before they were referred to our sleep clinic. A combination of advices and/or treatments was possible, see Table 3. Obvious, none of these were completely effective in these patients.
Medication, specifically melatonin, was tried out most often. In 16 out of 20 patients (80%) melatonin was partially effective. Alimenazine was partially effective in 4 out of 7 patients and promethazine in 1 out of 5. Seventeen parents or professional caregivers received education or general information on sleep and sleep problems. This was partially effective in 8 patients (47%). In 3 out of 8 patients psychological or behavioral treatment was partially effective. At last, homeopathic treatment was partially effective for 3 out of 10 patients.
According to our research criteria and information gathered by the Sleep Questionnaire 43 (83%, n = 52) patients had a severe sleep problem at referral to the sleep clinic, most of them had severe night waking problems (88%), followed by problems with early morning waking (33%) and settling problems (12%). Co-sleeping (insists on sleeping with somebody else) occurred 3 or more times a week in 15 patients (37%), all of them were living at home with their parents. Daytime sleepiness and bedwetting occurred 3 or more times a week in respectively 13 (35%) and 32 (82%) patients aged 5 yrs or older (n = 39).

DISCUSSION

It is remarkable that more than one third of the referred individuals already received melatonin. Our clinical experience is that in some patients although melatonin treatment was successful in the beginning, after a few months night waking problems returned. Because of a persisting positive influence on settling problems, melatonin treatment was continued. In some patients melatonin has a lasting positive effect on settling problems, but not on night waking problems. Further research to this phenomenon will be conducted.
The distribution of the level of ID was quite equal. This is not conform results from earlier research which found more sleep problems in children with more severe levels of ID than children with milder levels of ID[3]. Perhaps parents and professional caregivers of individuals with more severe levels of ID more often hold the belief that sleep problems are part of the disability and do not ask for advice or help than those of individuals with milder levels of ID. This may lead to our finding that all levels of ID were presented in our outpatient sleep clinic in the same quantity.

REFERENCES

[1] Wiggs L. Sleep disorders. In: Carr A, O'Reilly G, Noonan Walsh P, McEvoy J, eds. The handbook of intellectual disability and clinical psychology practice. East Sussex: Routledge, 2007: 371-421.
[2] Van der Heijden KB, Smits MG, Gunning WB. Sleep hygiene and actigraphically evaluated sleep characteristics in children with attention-deficit/hyperactivity disorder and chronic sleep onset insomnia. J Sleep Res 2006; 15:53-60.
[3] Wiggs L, Stores G. Severe sleep disturbance and daytime challenging behaviour in children with severe learning disabilities. JIDR 1996; 40: 518-528.
[4] Didden R, Korzilius H, Van Aperlo B, Van Overloop C, De Vries M. Sleep problems and daytime problem behaviours in children with intellectual disability. JIDR 2002; 46: 537-547.
[5] Didden R, Korzilius H, Smits MG, Curfs LMG. Sleep problems in individuals with Angelman syndrome. AJMR 2004; 109: 275-284.