Sleep disorders in Tourette syndrome in children and adolescents

Zaburzenia snu u dzieci i młodzieży z zespołem Tourette'a

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ABSTRACT

Introduction and aim of the study: The aim of study was comparison the sleep disorders in patients with TS, diagnosed in the Chair and Department of Developmental Neurology Poznan University of Medical Sciences Poland in 2005–2007 to the group of healthy children in similar age. **Material and methods:** The research was conducted on 81 patients with TS and the group of 156 healthy children. The ICSD-2 was the base of sleep disorder criteria using during anamnesis in both groups. **Results:** One or more sleep disorder appeared statistically more frequent in TS ($p<0.001$). Bruxism ($p<0.001$), somnambulism ($p<0.001$), snoring ($p=0,009$), bed wetting ($p=0,036$) were statistically more frequent in TS group. The prevalence of sleep talking ($p=0.492$), nightmares ($p=0.146$), sleep terror ($p=0.061$) was similar in both groups. **Conclusions:** Sleep disorders were more common in children with TS in comparison to control group. There is a need of additional research to evaluate influence of sleep disorders in children with TS.

Key words: Tourette syndrome, sleep disorders, children, adolescents.

STRESZCZENIE

Wstęp i cel pracy: Celem pracy było porównanie zaburzeń snu u dzieci z zespołem Tourette'a hospitalizowanych w Klinice Neurologii Wieku Rozwojowego Uniwersytetu Medycznego w Poznaniu w latach 2005-2007 z grupą kontrolną zdrowych dzieci w podobnym wieku. **Materiał i metody:** Badanie zostało przeprowadzone na 81 pacjentach z zespołem Tourette'a. W grupie kontrolnej było 156 zdrowych dzieci i młodzieży. Ankieta dotycząca zaburzeń snu, która badane były badane obie grupy dzieci została utworzona w oparciu kryteria zawarte w ICSD-2. **Wyniki:** Co najmniej jedno zaburzenie snu występowało statystycznie częściej w grupie dzieci z zespołem Tourette'a ($p<0.001$). Bruksizm ($p<0.001$), somnambulizm ($p<0.001$), chrapanie ($p=0,009$), moczenie nocne ($p=0.036$) pojawiało sie statystycznie istotnie częściej a grupie dzieci z zespołem Tourette'a. Częstość występowania mówienia przez sen ($p=0.492$), koszmarów sennych ($p=0.146$), lęków nocnych ($p=0.061$) była porównywalna w obu grupach pacjentów. **Wnioski:** Zaburzenia snu są częstsze u dzieci z zespołem Tourette'a w porównaniu do grupy kontrolnej. Konieczne są dalsze badania na większej grupie dzieci oceniające spektrum zaburzeń snu w zespole Tourette'a.

Słowa kluczowe: zespół Tourette'a, zaburzenia snu, dzieci, młodzież

TS (Tourette syndrome) is defined as a chronic, idiopathic syndrome with both motor and vocal tics beginning before adulthood. Tics are repetitive, stereotyped movements or vocalizations, which appear also in all of sleep stages in about 90% of patients with tic disorders and predispose to arousals and other sleep disorders [1-8]. Both in TS and sleep disorders there are dysfunction of serotoninergic, cholinergic and adrenergic neurotransmission what can cause concomitance both of disorders [3,7,9]. The two aminergic transmitters serotonin and andnoradrenaline have been found to be involved in sleep regulation as well as pathophysiology of Tourette's syndrome [3]. Severe course of TS correlates with more number of involuntary movements during the sleep and that is connected with more frequent arousals and sleep disorders [1,3,7]. Sleep disorders, especially parasomnias occur sporadically or periodically in about 14-47% of healthy children, and are observed in 20-60% patients with TS [3,7,10-15]. So general state of health seem to have very important influence on sleep but the etiology of sleep disorders are complex and it depends on varied factors connected with health, environment, family and social influences. Among NREM sleep parasomnias which are one of more frequent sleep disorders in children there are found sleepwalking, and sleep terrors [10,16,17]. Parasomnias as nightmares usually associated with REM sleep. Primary nocturnal enuresis is defined as involuntary micturition during the sleep more than two times a week in children over 5 years old [10,18]. Sleep related movement disorders as bruxizm are observed very frequent in children. According to ICSD–2 classification snoring and sleep talking belong to isolated symptoms, apparently normal variants and unresolved issues [10,11,19-22]. The aim of study was presentation frequency of appearance the most common sleep disorders in children with TS and comparison to the healthy control group in similar age.

MATERIALS AND METHODS

The research group consisted of 81 children and adolescents with TS hospitalized and diagnosed between 2005 – 2007

at Chair and Department of Developmental Neurology Poznan University of Medical Science in Poland. There were 16% (n=14) of girls and 84% (n=67) of boys between 6 and 18 years old, age mean was 11±3.1. The control group (CG) consisted of 156 children and teenagers from Poznan schools and kindergartens. CG was composed of 54% (n=85) of boys and 46% (n=71) of girls, between 4 and 18 years old age mean was 10±4.3, who at the moment of carrying the research were not diagnosed with neurological and mental disorders. The characteristic of the groups is shown in Table I. All patients with tic disorders fulfilled criteria for TS enclosed in DSM-IV-TR [23]. The criteria for including a patient into the TS group were verified during an interview with children, and also their parents or guardians and during the neurological test conducted by the authors. The criteria of sleep disorders enclosed in ICSD-2 was the base of sleep disorders evaluation during interview in both group of investigated children. Descriptive statistics were used to summarize the different kind of sleep disorders in TS group and CG. Mann Whitney U test was used to analyze the data for non-normally distributed continuous variables, and independence a chi-square test for categorical variables. Statistical analysis was completed using the computer statistical program Statistica 7.0.

Table I. Characteristic of in Tourette syndrome group and control group

	TS	CG
Number of children	n=81	n=156
Age (years)	11±3.1	10±4.3
Girls	n=14 (16%)	n=71 (46%)
Boys	n=67 (84%)	n=85 (54%)
M : F	4.7: 1	1.2: 1

RESULTS

The frequency of the following sleep disorders in group TS and CG were significantly different. In the case of the presence of at least one sleep disorder which appeared in 85% (n=69) of children in TS group and 63% (n=99) of children in CG group. The difference was statistically important (p<0.001). Bruxism was observed in 35% (n=28) of patients with TS and 15% (n=23) children, and it was significant differences (p<0.001). Somnambulism was noticed by the parents in 17% (n=14) of the patients with TS and 4% (n=7) of healthy children, and the differences was statistically important too (p<0.001). Snoring appeared in 35% (n=28) children with TS and 19% (n=30) in CG. This difference was significant (p=0.009). Primary bed wetting concerned 7% (n=6) patients with TS and 2% (n=3) healthy children, and it appeared statistically more frequent in TS than CG group (p=0.036). Sleep talking was present in 46% (n=37) patients in TS group and 41% (n=64) of children in CG, and the differences was not significant (p=0.492). Nightmares reported 16% (n=13) persons in TS group and 10% (n=15) healthy children. These were not statistically important differences too (p=0.146). The sleep terrors appeared in 10% (n=8) of children in TS group and 4% (n=6) healthy children and were not observed statistically more frequently in the TS group than CG (p=0.061). The frequency of appearance of one or more sleep disorders and the characteristic of particular sleep disorders in TS and CG group were shown in Table II.

Table II. The frequency of appearance sleep disorder in Tourette syndrome group and control group

	TS		CG		p
	n	%	n	%	
Sleep disorder (one or more)	69	85%	99	63%	<0.001**
Snoring	28	35%	30	19%	0.009*
Sleep talking	37	46%	64	41%	0.492
Somnambulism	14	17%	7	4%	<0.001**
Bruxism	28	35%	23	15%	<0.001**
Sleep terror	8	10%	6	4%	0.061
Nightmares	13	16%	15	10%	0.146
Bed wetting	6	7%	3	2%	0.036*

* p< 0.05; ** p<0.001

DISCUSSION

Sleep disorders have important influences on proper social functioning of children especially with diagnosis of TS. Somnambulism, bruxizm, sleep talking, nightmares, sleep terrors belong to sleep disorders which most often appeared in children especially with diagnosis of TS [1,6,15,24,25]. At least one sleep disorder in the TS group was in upper limit of frequency described by other authors i.e. more than one sleep problem appeared in 85% of children in the TS group. In CG percentage of children with one or more sleep disorders exceeded upper limit of data quoted by others i.e. they appeared in 63% of children in CG group. Probably this occurrence was caused by reporting almost all also episodically noticed sleep disorders by parents of healthy children [26-28]. But even in this case the differences between both investigated groups were statistically important (p<0.001).

Snoring often appears in children and adolescents and it can be serious problem of lower sleep quality and sleepiness during the day and this is one of the more important factor of concentration and attention deficits [2,16,21,29]. The frequency of snoring in healthy children population is evaluated on 7 – 23% [16,29-31], but in the literature there are no detailed data concerning the prevalence of this problem in patients with TS. However it's known that snoring is observed more frequently among children with TS [4,5,24]. Snoring predisposes to other parasomnias as sleep talking, grinding of teeth or nightmares [16,29]. The parents of 19% children in CG noticed this problem, and

the prevalence was similar to other authors' data [16,29-31]. Snoring was noticed almost twice more often in the TS group i.e. in 35% of children. This difference was statistically important (p=0.009).

Bruxism is defined as grinding the teeth or a clenching of the teeth, which is associated with forceful jaw movements during sleep. This phenomena appeared most often in the first part of night, in NREM sleep, but it can occur in all sleep stages [32]. The most frequent consequences of bruxism is headache, wearing and breaking of teeth, hypersensitivity of teeth and other problems connected with dysfunction of stomatognathic system [33]. Bruxism appeared in about 9 - 25% between 3 and 17 years old [10,11,30-32,34] but in the literature there are no data concerning frequency of bruxism appearance in children with TS. In many cases, no treatment is necessary, but sometimes behavioral therapy, protective dental covers or psychotherapy is helpful. Teeth grinding was noticed in 15% of healthy children and this prevalence was similar to the data from literature [11,30-32,34]. Frequency of bruxism in children with TS was observed statistically more often than in CG (p<0.001).

Somnambulism is a disorder of arousal which appears in 3 and 4 stage of sleep NREM. Motor activity during the episode of somnambulism seems to be reasonable, but nothing is remembered [10,13,35]. Sleep walking is observed in 6 – 29% of healthy children between 8 and 12 years old [11,19,30,31,34]. In almost all cases no treatment is necessary [32]. Pharmacotherapy can be indicated in very frequent episodes of somnambulism which are life or health hazards [32]. Somnambulism is often accompanied by tic disorders and the prevalence of that parasomnia in patients with TS concern about 17 – 41% [10,36,37]. In our study, 4% of healthy children and 17% of patients with TS had episodes of somnambulism. The percentages of children with sleep walking in both groups were similar to the data quoted in literature [11,19,30,31,34]. The differences between these groups were statistically important (p<0.001).

Primary enuresis nocturna is not connected with any sleep stages, it concerns especially younger children in who excluded organic causes of bed wetting, and the prevalence of this sleep disorder decreases in conjunction with age [10,11,14,18,30,31,34]. Enuresis nocturna appears more frequently and lasts longer in patients with TS than in healthy children of similar age [18,25,34]. In our study percentage of children in both groups were lower than the quoted in literature, but the difference between groups were statistically important (p=0.036). As bed wetting is still an embarrassing problem in Polish society, the results can be lower than the real life prevalence of this sleep disorder.

Sleep talking or somniloquy occurs when children talk loudly during sleep, but listeners may or may not be able to understand what is said [12]. It appears frequently in 4 - 14% of healthy children, and occasionally in 22 – 60% of that population [10,11,19,20,30,31]. Frequency of this phenomenon is lower in conjunction with age. Sleep talking is statistically more frequent in children with TS in comparison with healthy children [15]. In our study there are no significant differences between CG and TS group (p=0.492). Probably the result was caused by more careful inter alia observation of sleep children with tic disorders by parents.

Night terrors are parasomnia which appear in healthy children in the first three hours of sleep, during stage 3 and stage 4 NREM sleep [10,12,32]. Night terrors are recurrent episodes of crying and fear with activation of autonomic system which last several minutes [16,30,31]. Children do not recall a dream after a night terror and do not remember anything next morning [14]. Night terrors are closely connected with age and are most common among children aged 3 – 12 [11,20]. An estimated 3 – 15% of healthy children experience night terrors during the sleep [11,19,20,34]. The most important way of treatment is to educate the family about the disorder and the reassurance that the episodes are not harmful for children [12,32]. It is known that night terrors are more frequent in children with tic disorders [25,36,37]. The prevalence of them in children with TS is about 10 – 16% [25,36]. Nightmares appear quite regular in 4 – 6% of children, but occasionally mention them 9 – 17% of children [11,20,30-32]. Nightmares are repeated episodes of a frightening, unpleasant dream that appears in REM sleep. In the morning the child remembers contents and fear accompanying the dream. Nightmares usually are caused by strong, unpleasant emotions [32] but also by the treatment of some drugs as benzodiazepines, antidepressants, barbiturates, or the withdrawal of them [12,32,38]. Medications are neither helpful nor indicated but psychotherapy in some cases can be beneficial [12,39,40]. Unpleasant dreams appear in 15-40% children with TS [15]. In our study there were no significant differences concerning sleep terrors (p=0.061) and nightmares (p=0.146) between CG and TS. Although there was tendency to occurrence more frequent both of parasomnias in children with TS.

CONCLUSIONS

Parasomnias were observed such as bruxizm, somnambulism, snoring, enuresis nocturna were observed statistically more frequent in children with TS in comparison to healthy children from CG. Presence of motor and vocal tics while sleeping are one of the most important reason of arousals because they increase the probability of mentioned sleep disorders among children and adolescents with TS. So general state of health have very important influence on quality of sleep but the etiology of sleep disorders are complex and it depends on varied factors connected also with environment, family and social influences. There is a need of additional research to evaluate influence of sleep disorders on natural course of Tourette syndrome.

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