

Disturbed sleep in children with Tourette syndrome A polysomnographic study

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Abstract

Objective: To evaluate objective data on sleep quantity/quality and motor activity during night sleep in children with Tourette syndrome (TS). **Method:** Polysomnography of 17 unmedicated TS children (ages: 7;11–15;5, mean: 11;10 years) without comorbid attention-deficit hyperactivity disorder (ADHD) was compared with 16 age-, sex- and IQ-matched healthy controls. Sleep analyses according to the procedure of Rechtschaffen and Kales were supplemented by counting epochs with short arousal-related movements (≤ 15 s), thus allowing to calculate correlations between motor activity and sleep parameters. **Results:** Children with TS demonstrated changes in sleep parameters, including longer sleep period time, longer sleep latency, reduced sleep efficiency, and prolonged wakefulness after sleep onset. Their sleep profiles showed significantly more

time awake and less sleep stage II. However, REM sleep variables, slow-wave sleep, and number of sleep stage changes were unaffected. Movement time was similar in both groups, but epochs with short arousal-related movements were increased in TS. Further analyses showed no significant correlations between sleep parameters and nighttime nontic movements, level of psychopathology or tic severity during daytime. Periodic limb movements during sleep (PLMS) were only seen in one TS patient (low PLMS index of 7.8/h). **Conclusions:** Children with TS have disturbed sleep quality with increased arousal phenomena, which both may be intrinsic to the disorder and might trigger tics and other behavioral problems during daytime. This indicates the need for sleep evaluation in patients with TS. © 2003 Elsevier Inc. All rights reserved.

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Introduction

A variety of behavioral disturbances often associated with Tourette syndrome (TS) include attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder, learning disabilities, anxiety, depression, and self-injurious behavior [10,27,37]. Several studies have also shown that the night sleep of children and adolescents with TS seems to be markedly disturbed. Observations by caregivers indicated sleep problems in 20–50% of children and young adult patients with TS. Difficulties in falling and staying asleep, separation anxiety in the evening, and

parasomnias (sleep walking and pavor nocturnus) were the most common problems [4–6,14,19,20,25,30,34,38,44]. In cases of a positive family history concerning tics, sleep disturbances of children and adolescents were even more frequent [21,23,34]. Associated ADHD as well as migraine may also lead to an increase of sleep problems in TS patients [4,6,14].

Aiming to gather objective data concerning sleep behavior of TS patients, some studies have used video- and polygraphic measurement systems. The few existing polysomnographic studies of children and adolescents with TS have almost exclusively reported sleep EEG findings of mixed samples including both children and adults [18,21,28,42], while Cohrs et al. [12] investigated only adult patients with TS. Moreover, studies differed with respect to group size (those with less than five TS patients

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are not mentioned in this overview), the setting (sleep laboratory vs. assessment at home), number of registered nights (1–4), and reference data (healthy controls vs. normative data taken from previously published reports), thus making comparisons between studies difficult.

In summary, sleep EEG findings of TS patients show many problems (review in Refs. [26,39]). In general, REM sleep might be decreased and slow-wave sleep enhanced as compared to healthy controls. But these findings are inconclusive. Sleep efficiency of TS patients may be diminished due to their prolonged wakefulness after sleep onset, while sleep apnea, tics, and frequent arousal reactions may additionally disturb sleeping behavior. Some indirect evidence points to more frequent parasomnias in younger TS patients. Given the very small samples studied and their wide age range as well as uncontrolled EEG disturbances while awake [21], the validity of previous polysomnographic findings seems limited. Furthermore, the results obtained by behavioral assessment of the nocturnal sleep quality of children and adolescents with TS (difficulties in falling and staying asleep, separation anxiety, somnambulism, pavor nocturnus, and nightmares) have not received clear confirmation in polysomnographic studies.

Thus, the available data on sleep behavior of children and adolescents with TS is scarce and arbitrary and points to further polysomnographical investigations with updated methodology to objectively evaluate the sleep behavior of children and adolescents with TS as a separate age group, since sleep disturbances can have negative effects on their cognitive and emotional status and thus increase preexisting problems of their daytime behavior [46]. The objective assessment of the specific nature and frequency of sleep disturbances in children with TS is important in order to optimize treatment methods for TS. Since nontic motor phenomena during sleep seem to be increased in TS patients [12] and some adult patients with TS might show periodic limb movements (PLMS [33,48]), it would also be of particular interest to evaluate sleep behavior with respect to nontic movements.

Therefore, the following assumptions should be tested by polysomnography: (1) compared to healthy controls, children with TS may show qualitative sleep deficits, with decreased sleep efficiency, prolonged sleep latency, increased wakefulness after sleep onset, increased number of awakenings, while there might be unpredictable changes in slow wave sleep and REM sleep. Moreover, (2) there may be more nontic body movements and enhanced occurrence of motor arousal during all sleep stages in TS patients compared to control subjects, and some children with TS might show PLMS.

Methods

Seventeen children and adolescents with TS and a group of 16 healthy controls matched for age, gender, and non-

verbal IQ were studied by polysomnography on two sequential nights.

All patients with TS were consecutive referrals to the Department of Child and Adolescent Psychiatry at the University of Göttingen. Children and adolescents with TS were included in the study after being clinically examined independently by two board certified child psychiatrists and fulfilled the inclusion but no exclusion criteria as described below. Fourteen of the 17 patients were referred to the sleep laboratory from an outpatient setting, while 3 patients were recruited from the in-patient unit of the clinic. Healthy controls, recruited among friends and relatives of the clinical staff, had no psychiatric, neurological, or internal disorders.

The study was approved by the local ethics committee of the Medical Faculty of the University of Göttingen and was performed in accordance with the ethical standards of the Declaration of Helsinki. A detailed description of the study was provided to the parents and their children. The parents of each subject gave written consent and the children age-appropriate consent after complete information about the study.

All patients fulfilled the diagnostic criteria for TS according to the International Classification of Diseases (F95.2, ICD-10; WHO, 1992) and the Diagnostic and Statistical Manual of Mental Disorders (307.23, DSM-IV; APA, 1994). Subjects with a diagnosis of a hyperkinetic disorder or ADHD following the ICD-10 and the DSM-IV criteria, as well as patients with other psychiatric (obsessive-compulsive disorder, pervasive developmental disorder, autism, psychosis, posttraumatic stress disorder, or primary anxiety or affective disorder), neurological (including brain injury or relevant problems like epileptiform activity in EEG), or internal medical impairment were excluded. Subjects (patients and controls) were not included in the study if their Verbal IQ and Performance IQ were both less than 80, as assessed by the German version of the WISC-R [47]. In order to avoid potential selection biases, prestudy sleep behavior was not a selection criterion for participation in the study.

A semistructured clinical interview with parents and patients as well as clinical observations of each subject, including physical examination, served as basis for diagnosing the psychiatric status and especially to guarantee exclusion of comorbidity with ADHD. Psychopathological questionnaires [1,7,11,22,45,49] were administered. Mean severity of TS as rated on the TS Severity Scale (TSSS [40]) was 3.4 (S.D.: 1.8) on average, indicating a moderate severity of TS. The raters of TS severity were blind to the polysomnographic data.

For assessment of sleep quality at home, parent reports were obtained with the Child Behavior Checklist (CBCL) [1]. The CBCL is a widely used instrument that assesses dimensions of childhood psychopathology as observed during the last 6 months. There are 113 items that are rated as being “not,” “somewhat,” or “very true,” which are combined to

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