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Childhood parasomnia – A disorder of sleep maturation?

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ABSTRACT

Background: Childhood parasomnias are believed to be a benign disorder due to immaturity of some neural circuits, synapses and receptors. The aim of our study was to explore a possible connection with other neurological developmental disorders.
Methods: 72 children (mean age 9.9 ± 5.0 years, 47 boys) were clinically examined and 88 nocturnal v-PSG and 22 v-EEG recordings were evaluated. The most frequent diagnostic findings were: sleepwalking in 24 children, confusional arousal in 21, sleep terror in 8, groaning and enuresis each in 7, non-specific arousal disorder in 4 patients, and REM-related parasomnia in only one child. For statistical evaluation chi-square test, the two-sample t-test and Mann–Whitney rank test were used.
Results: Perinatal risk history was found in 38% of the cohort. Developmental disorders were diagnosed in 30 children (41.7%), more frequently in combinations with: attention-hyperactivity disorder (30.6%), dyslexia and dysgraphia (13.9%), developmental dysphasia (9.7%), mild motor and/or intellectual dysfunction (6.9%). Abnormal movements in sleep, some of them also regarded as developmental, were diagnosed in 37 children (51.4%). Sleep-related breathing disorders were found in 29 patients (40.3%) –snoring (29.2%) and/or sleep apnea (11.1%). Only 16.7% had no comorbidity. Most of the children (60%) showed 2 or 3, exceptionally up to 5 comorbidities. Children, in whom no parasomnia was found in close relatives, had a mild but non-significant earlier onset of the disease (4.4 ± 4.0 against 6.3 ± 4.3 years).
Conclusion: Childhood parasomnias are frequently associated with perinatal risk factors and developmental comorbidities, and can be regarded as a disorder of sleep maturation.

1. Introduction

Parasomnias are undesirable events that accompany sleep and can occur while falling to sleep, during sleep, or during arousals from sleep. According to the international classification of sleep disorders,[1] three main groups are distinguished: (1) disorders of arousal arising from NREM (Non-Rapid Eye Movement) sleep, (2) parasomnias usually

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associated with REM (Rapid Eye Movement) sleep, and (3) other parasomnias occurring in different sleep stages.

Arousal disorders are seen as benign phenomena, especially in children. The common features are: onset in childhood with cessation in adolescence, presence of triggering factors (sleep deprivation, fever), occurrence in the first third of the night with awakening from deep delta sleep, long duration of each episode occurring only once in the same night, minimal recall of the event, and strong familial pattern.[2] Three basic types are differentiated: confusional arousals, sleepwalking (somnambulism) and sleep terrors. All parasomnia attacks may take minutes up to half an hour to pass. They are marked by poor arousability, confusion, poor coordination, and by a tendency toward aggressive response to waking attempts. More than one type may coexist in the same patient; as an expression of the same pathophysiological continuum.[3]

The main features of confusional arousals include disorientation with confusional behavior and relative unresponsiveness to environmental stimuli.[4] An episode of sleep terror usually begins with vocalization, sudden sitting up in bed, screaming, crying, and showing facial signs of intense fear with autonomous system activation (tachycardia, tachypnea, mydriasis, sweating, reddening).[5] Sleepwalking consists in recurrent episodes in which the subject, without awakening, exhibits complex automatic behaviors with leaving the bed and walking some distance.[6] Some episodes of arousal parasomnias may mimic epileptic attacks, particularly seizures originating from the frontal lobe.[2,7,8]

The most common childhood REM parasomnias are nightmares, less frequently sleep paralysis and REM behavior disorder. Nightmares are terrifying dreams leading to prompt awakening, usually in the second half of the night, with a clear recollection of the dreams.[6] Sleep paralysis is marked by episodes of inability to perform voluntary movements either at sleep onset or on awakening; REM behavior disorder is characterized by intermittent absence of the normal REM sleep atonia, and associated with intense motor activity related to dream mentation.[9]

The most frequent parasomnias occurring in both - NREM and REM sleep – include nocturnal enuresis, defined as recurrent involuntary voiding in sleep after the age of five years, and groaning (catathrenia), characterized by prolonged expiration with loud monotonous vocalization.[10] In the past decades some studies demonstrated a close connection between arousal secondary to nocturnal breathing disorders – sleep apnea and hypopnea.[11,12] A close connection between parasomnias and sleep-related movement disorders is well known, too.[13] Some of them – e.g. rhythmic movement disorder and bruxism – are physiological in infants and toddlers. Most will resolve by the age of 3–4 years, suggesting a close ontogenetic relation with early-age parasomnias.

Childhood parasomnias are believed to be a benign disorder caused by immaturity of some neural circuits, synapses and receptors.[14] Most of them are likely to disappear during adolescence. Our aim was to evaluate a possible association between parasomnias and developmental disorders in a cohort of children referred to us for neurological and sleep disorder treatment.

2. Patients and methods

Seventy-two children and adolescents with parasomnia (47 boys, 25 girls, mean age 9.9 ± 5.0 years, age range 2–19 years) were examined in a cross-sectional study during the period 2005–2011. All data were retrospectively processed. All subjects were clinically examined, and their parents underwent clinical face-to face interviewing by one of the authors (SN, IP, DK) for information about perinatal risk, mild motor and/or intellectual development dysfunction, speech disorder and learning disabilities, attention deficit and hyperactivity disorder (ADHD). They were asked to describe the child's nocturnal episodes, their frequency, severity, age at onset and any possible combination with sleep disorder breathing and/or sleep related movement disorders. Clinical testing took the form of neurological, in most patients neuropsychological, and in part of them, psychiatric and phoniatric examinations. A total of 88 nocturnal video-polysomnographic (v-PSG), and 22 video-electroencephalographic (v-EEG) recordings were evaluated in 72 patients. V-PSG records as well as v-EEG were standardized as regards the time schedule and adapted to childrens' habits (usually 9.30 p.m. up to 6.30 a.m.), and so was the standard-connection equipment (Schwarzer polygraph). PSG records were evaluated visually according to Rechtschaffen and Kales[15] and American Academy Sleep Medicine manual[16,17] rules and, similarly as EEG recordings, evaluated by two neurophysiologists (IP, DK). The diagnosis was verified in 55 cases (76.4%) by clinical episodes during the nocturnal monitoring, in 9 cases (12.5%) by prolonged awakening from the delta sleep – typical of arousal parasomnias. Only 8 cases (11.1%) were diagnosed solely according to clinical features as reported by childrens' parents.

In the study period, the most frequent diagnosis was sleepwalking in 24 patients, confusional arousal in 21, sleep terror in 8, groaning and enuresis each in 7, non-specific arousal disorder in 4 cases, and REM-related parasomnia in one child. Mean age at the beginning of episodes was 4.9 ± 4.3 years.

For statistical evaluation (JS) chi-square test, the two-sample t-test and Mann–Whitney rank test were used. The results were expressed as a mean ± SD, where a p-value of less than 0.05 was considered significant. The statistical analysis was made using BDMP Statistical Software, release 8.1 (Cork, Technology Park, Ireland).

The study was approved by the Ethics Committee and made to conform to the latest guidelines of the Declaration of Helsinki. Informed consent was obtained from all of the subjects' parents. There is no potential financial, professional or personal conflict that would be relevant to the manuscript.

3. Results

Perinatal risk factors including abnormal and/or maintained pregnancy, premature delivery, asphyxia and other post-delivery risks were found in more than one third of the cohort (37.7%). Mild motor and/or intellectual developmental impairment was diagnosed in 5 children, developmental dysphasia (specific speech disability) in 7 cases, 10 children