Rhythmic Movement Disorder in Sleep Persisting into Childhood and Adulthood

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**Study Objectives:** To evaluate the type, duration, and distribution of rhythmic movements in sleep stages in school-aged children and young adults; to find out if cases of rhythmic movement disorder persisting beyond infancy are associated with any daytime symptoms or psychopathology.

**Design:** All participants underwent neurologic examination, biochemical screening, electroencephalography, neuroimaging, overnight videopolysomnography, and psychologic examination.

**Setting:** Department of Neurology and Sleep Laboratory, 1st Medical Faculty, Charles University, Prague

**Patients or Participants:** Ten subjects referred to the sleep disorders center because of rhythmic movement disorder. Five males, 5 females; age range, 7-24 years; mean age 14.7 ± 5.69 years.

**Interventions:** None.

**Measurements and Results:** Biochemical screening, electroencephalogram, and neuroimaging were unremarkable in all cases. According to duration, 2 types of rhythmic movements were observed: polysomnography: longer episodes appeared in wakefulness and in non-rapid eye movement stage 1 sleep, while shorter episodes (2-80 seconds) occurred during non-rapid eye movement stage 2, non-rapid eye movement stage 3-4, and rapid eye movement sleep. According to sleep-stage distribution, we defined (a) rhythmic movements prevailing in the first half of the night and in the morning hours, usually associated with wakefulness or superficial sleep; (b) rhythmic movements occurring throughout the night in all sleep stages; (c) rhythmic movements prevailing in the second half of the night and mainly associated with rapid eye movement sleep. Psychologic examination showed symptoms of the attention-deficit/hyperactivity disorder in 6 cases.

**Conclusions:** According to our study, rhythmic movement disorder persisting beyond infancy may be connected with various daytime symptoms; a strong association between rhythmic movement disorder and attention-deficit/hyperactivity disorder was found in school-aged children. We speculate that pathogenetic mechanisms similar to those in attention-deficit/hyperactivity disorder are involved in rhythmic movement disorder or that symptoms of attention-deficit/hyperactivity disorder may be secondary to rhythmic movement disorder.

**Keywords:** Rhythmic movement disorder, sleep architecture, polysomnography, attention-deficit/hyperactivity disorder

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**INTRODUCTION**

RHYTHMIC MOVEMENT DISORDER (RMD) DURING SLEEP IS, ACCORDING TO THE INTERNATIONAL CLASSIFICATION OF SLEEP DISORDERS, DEFINED AS A GROUP of stereotyped repetitive movements involving large muscles usually of the head and neck that typically occur prior to sleep onset and are sustained into light sleep.1-4 RMD is observed in 9-month-old infants at a rate of up to 66%, decreasing dramatically to the age of 4 years (8%).

Seen as a benign condition, it is likely to resolve in preschool-aged children.5-7 In rare cases (particularly those associated with mental retardation), violent movements can cause head or eye injuries.5,6 In infancy, rhythmic movements are suspected to be a learned “self-stimulating” or “autoerotic” behavior connected with pleasurable feelings, apparently a tension-releasing maneuver of considerable hypnogenic potential.10,11 Other hypotheses refer to the maturational role of rhythmic movements in early psychomotor development by means of vestibular apparatus stimulation and to a connection with central nervous system rhythmicity.10-13

In the past decades, sporadic cases of RMD have been described that do not meet the criteria of the sleep-wake transition parasomnia as defined by International Classification of Sleep Disorders. The clinical picture of RMD is the same, but movements persist in all sleep stages, possibly even prevailing in rapid eye movement (REM) or slow-wave sleep.15-28 In such cases, RMD persists from early childhood until school age or even adulthood. It may underlie a major psychosocial handicap and even daytime symptoms.11,21-23 In these cases, however, RMD is not associated with significant mental retardation, autism, or other severe psychiatric disorders, as has been previously thought. Pathophysiologic factors causing the incorporation of rhythmic movements into different sleep stages remain obscure.

The aims of our study were to evaluate the type, duration, and distribution of rhythmic movements in sleep stages in school-aged children and young adults and to find out if cases of RMD persisting beyond infancy are associated with any daytime symptoms or psychopathology.

**SUBJECTS AND METHODS**

In a 6-year period, we evaluated 10 subjects (5 male, 5 female) aged 7 to 24 years (mean age 14.7 ± 5.69 years) with RMD persisting from infancy and occurring daily during different parts of the night. The follow-up period ranged from 2 months to 6 years.

During the period, all patients underwent neurologic examination, biochemical tests, neuroimaging (computed tomography or magnetic resonance imaging of the brain), electroencephalography (EEG), nocturnal video-polysomnography, and psychologic examination. All patients or their parents were asked to sign an

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informed consent for neuroimaging examination.

Standard nocturnal videopolysomnography was performed in all cases. The number of nights of recording depended on the occurrence of rhythmic movements (2-3 consecutive nights).

A Schwarzer polygraph was used for all polysomnography studies with standard EEG montage (F8-T4, T4-T6, F4-C4, C4-P4, F3-C3, C3-P3, F7-T3, T3-T5, T6-C3, T5-C4), submental electromyography, and horizontal electrooculography. Electrocardiogram, bilateral anterior tibialis electromyogram, and videorecording using an infrared-light camera were also studied. The respiratory air flow, effort parameters, and transcutaneous oxygen saturation were monitored. Respiratory parameters were evaluated with standard techniques. Neither esophageal pressure measurement nor nasal cannula/pressure transducer was used for technical reasons. Sleep stages were visually scored according to the Rechtschaffen and Kales standard criteria in 30-second epochs.29

Sleep-stage scoring during rhythmic movements was difficult because of pervasive motor artifacts in all polygraphic channels. If a rhythmic-movement episode lasted more than a half an epoch, precluding sleep-stage identification, or if it extended over more epochs, it was scored as movement time: if sleep preceded and followed, it was scored according to the standard scoring rules. It was also classified as movement time if sleep only preceded and if the rhythmic-movement episode was followed by wakefulness. This was because it was impossible to tell at exactly which time vigilance appeared in rhythmic movements. If the rhythmic-movement episode was shorter than an epoch, the epoch was scored according to the apparent sleep stage.

The duration, the number of rhythmic-movement periods, and their relation to sleep stages were measured in each patient separately. Each episode was classified according to the stage marking the onset of episode.

Psychologic examination included for Children-III and Wechsler Adult Intelligence Scale-Revised, drawing tests, Children’s Manifest Anxiety Scale, Trail Making Test, Complex Figure Test, and a direct interview.

RESULTS

Historical Data

Two patients had a positive family history of RMD. Five had a positive perinatal history (high-risk pregnancy and prematurity in 3 cases, hyperbilirubinaemia with phototherapy in 2 cases). Psychomotor development was generally normal except in 1 patient with slightly retarded development. Medical history revealed allergic symptoms in 7 subjects.

The age at rhythmic-movement onset varied from 0.5 to 3 years (in 2 children after hospitalization without parents).

At the time of investigation, the reported types of movements were headbanging (4 patients); bodyrolling (2); headbanging and headrolling (1); headrolling and bodyrolling (1); headbanging and bodyrolling (1); and headbanging, headrolling, and bodyrolling (1).

The movements were present almost every night. Based on parents’ reports, 6 patients suffered from the movements at sleep onset as well as in the second half of the night. In 4 patients, rhythmic movements occurred mostly in the second half of the night. Three patients also experienced occasional rhythmic movements during the day. Rhythmic-movement episodes lasted from a few up to 30 minutes, with a tendency to recur several times during the night. The children were fully asleep and difficult to wake up; a partial and temporary abatement of rhythmic movements was reported on being forced to change their body position, being touched, or hearing a loud sound. Five patients were partly aware of the movements.

One patient described a strong urge to behave like this before falling asleep and had feelings of aggression when he was prevented from it. He tried to alleviate rhythmic movements by moving his legs instead.

All parents of school-aged children complained of being disturbed in sleep by the children’s potentially injurious violent noisy movements. Adolescent and adult patients felt uncomfortable having to sleep in a room shared with other people. Other sleep disturbances were apparent in the history of 3 patients: parasomnia in 2 and difficulty in falling asleep in 1.

Parental and subjective reports showed various daytime complaints in all patients: attention difficulties (n, 7), hyperactivity (5), neurotic symptoms (5), tiredness and sleepiness during the day (4), morning headache (5), and compulsive behaviour (1).

Neurological and Psychologic Examination

Neurological investigation was normal in all subjects. Biochemical tests were unremarkable, including iron, transferrin, and ferritin levels. Brain imaging (computed tomography in 8, magnetic resonance imaging in 2 patients) revealed no pathologic findings. Electroencephalogram was normal in 6 patients, and 4 had nonspecific abnormalities (in 3 cases, there was irregular background activity with slow waves; in 1 case, diffuse theta activity).

On applying the criteria of the Diagnostic and Statistical Manual of Mental Disorders-IV,30 4 patients fulfilled the criteria for the attention-deficit/hyperactivity disorder of the inattentive type (ADHD-I), and 2 patients for the combined type of ADHD (ADHD-C).

Psychologic examination revealed depression in 1 patient and obsessive-compulsive disorder in another 1. Neurotic symptoms were present in 6, and specific learning difficulties in 3 cases (dyslexia in 2 cases, dysgraphia in 1 case). Neurotic symptoms included increased anxiety in 3; mood disturbances in 2, affective lability in 2; onychophagia in 2; and stress-related difficulties in falling asleep, intrapsychic tension, anxiety with prominent vegetative symptoms, and tics in 1 case each.

Intellectual level was within the normal range in 8 patients and borderline in 2 patients.

Nocturnal Videopolysomnography

Several types of rhythmic movements were recorded during overnight videopolysomnography (Table 1). Headbanging was as frequent as bodyrolling (both in 5 cases), headbanging was found in 2 cases. Two patients exhibited 2 types of rhythmic movements (headbanging and bodyrolling, headbanging and bodyrolling). In REM sleep, rolling movements alone were observed. The frequency of rhythmic movements varied from 0.5 Hz to 2 Hz, with 1 Hz as the prevailing frequency. The reported occurrence of rhythmic movements during the night was much greater under home conditions, as were combinations of various rhythmic-movement types. Rhythmic movements were reported to be present in all patients almost every night at home, but on the night of polysomnography, they appeared during the first night in

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5 patients, during the second in 2 patients, or even during the third night of monitoring in 3 patients.

The distribution of rhythmic movements according to the part of the night and sleep stages varied (Table 2). Rhythmic movements were observed in wakefulness before falling asleep, during the night, or in the morning hours in 7 patients; in non-REM (NREM) stage 1 sleep in 7 patients; in NREM stage 2 sleep in 6 patients; in NREM stage 3/4 sleep in 3 patients; and in REM sleep in 5 patients.

According to the distribution of rhythmic movements, we defined 3 groups: (1) patients 2, 4, 5, 6, 7, 9, who showed rhythmic movements prevailing on sleep onset, in the first half of the night, and in the early-morning hours—usually associated with wakefulness or superficial sleep (patients 2 and 4 showed sporadic rhythmic movements in slow-wave sleep or in REM sleep); (2) patients 1 and 8 showed rhythmic movements throughout the night in all sleep stages (Figure 1); and (3) patients 3 and 10 showed rhythmic movements prevailing in the second half of the night, mostly in REM sleep (Figure 2).

Total time spent during the night in rhythmic-movement epi-

Table 1—Overview of Patients

<table>
<thead>
<tr>
<th>Pts</th>
<th>Age, y</th>
<th>Sex</th>
<th>Age of onset, y</th>
<th>Type of Rhythmic Movement</th>
<th>Sleep Stage*</th>
<th>Results of Psychologic Examination</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>14</td>
<td>M</td>
<td>2</td>
<td>BR</td>
<td>W; NREM 1, 2, 3/4; REM</td>
<td>ADHD-I, neurotic symptoms, dysgraphia</td>
</tr>
<tr>
<td>2</td>
<td>24</td>
<td>M</td>
<td>2</td>
<td>HB</td>
<td>W; NREM 1, 2, 3/4</td>
<td>Depression, neurotic symptoms</td>
</tr>
<tr>
<td>3</td>
<td>7</td>
<td>F</td>
<td>1.5</td>
<td>HB, BR</td>
<td>W; NREM 1, 2; REM</td>
<td>Normal</td>
</tr>
<tr>
<td>4</td>
<td>14</td>
<td>F</td>
<td>1</td>
<td>HB</td>
<td>W; NREM 1</td>
<td>ADHD-I, dyslexia</td>
</tr>
<tr>
<td>5</td>
<td>13</td>
<td>F</td>
<td>2</td>
<td>HB</td>
<td>W</td>
<td>ADHD-I, dyslexia</td>
</tr>
<tr>
<td>6</td>
<td>14</td>
<td>M</td>
<td>3</td>
<td>HB</td>
<td>W</td>
<td>ADHD-C, neurotic symptoms</td>
</tr>
<tr>
<td>7</td>
<td>9</td>
<td>M</td>
<td>3</td>
<td>HB</td>
<td>W</td>
<td>ADHD-C, neurotic symptoms</td>
</tr>
<tr>
<td>8</td>
<td>10</td>
<td>F</td>
<td>1</td>
<td>HB, BR</td>
<td>W; NREM 1, 2, 3/4; REM</td>
<td>Obsessive-compulsive disorder</td>
</tr>
<tr>
<td>9</td>
<td>19</td>
<td>M</td>
<td>1</td>
<td>BR</td>
<td>NREM 1, 2</td>
<td>Neurotic symptoms</td>
</tr>
<tr>
<td>10</td>
<td>23</td>
<td>F</td>
<td>0.5</td>
<td>BR</td>
<td>REM</td>
<td>Neurotic symptoms</td>
</tr>
</tbody>
</table>

*Sleep stage in which rhythmic movements occurred.
Pt refers to patient number; HB, headbanging; HR, headrolling; BR, bodyrolling; W, wakefulness; NREM, non-rapid eye movement sleep; REM, rapid eye movement sleep; ADHD-I, attention-deficit/hyperactivity disorder of the inattentive type; ADHD-C, ADHD of the combined type.

Figure 1—Rhythmic movements in slow-wave sleep.
sodes ranged from 6 seconds to 85 minutes. Longer episodes appeared in wakefulness and NREM stage 1 sleep, while shorter episodes (2-80 seconds) occurred mostly during NREM stage 2, NREM stage 3/4, and REM sleep. Tending to occur in clusters, these shorter episodes were mostly neither preceded nor followed by arousal, as defined by American Sleep Disorders Association.\textsuperscript{31}

Some of the rhythmic-movement episodes during REM sleep were preceded and followed by loss of muscle atonia (preceding rhythmic movements by 30-60 seconds and lasting another 30-50 seconds afterward) in 3 patients (patients 3, 8, 10). In 1 patient (patient 5), this short loss of muscle atonia was found without any rhythmic movements. (Figure 3)

The usual sleep parameters were analyzed (Table 3). In 3 patients (patients 2, 6, 9), sleep fragmentation with frequent arousals from all sleep stages was evident without any connection with rhythmic movements.

Polysomnography also showed repeated arousals from delta-wave sleep, suggestive of parasomnia, in 3 patients; bruxism in 2 patients; somniloquy in 3 patients; ronchopathy in 2 patients; slight obstructive apnea syndrome in 1 patient; epileptic discharges during NREM stage 2 sleep in 2 patients; and short and nonperiodic leg movements in 2 cases. There were no significant respiratory, cardiovascular, or EEG changes preceding rhythmic movements. During rhythmic movements, tachycardia was observed in most cases. As neither esophageal pressure measurement nor nasal cannula/pressure transducer was used, we cannot exclude subtle breathing-pattern changes.

**DISCUSSION**

Recent reports support the idea that not only RMD is a type of parasomnia of early childhood likely to resolve in preschool age, but that in some cases it lasts long into school age, adolescence, or adulthood in a rather troublesome fashion. In these cases, rhythmic movements not only are associated with superficial sleep stages during falling asleep, but will occur in any sleep stage through the night,\textsuperscript{11,14-28} as we also observed in some of our patients. These findings raise the question of whether rhythmic movements of infancy and persistent rhythmic movements belong in the same pathophysiologic category.\textsuperscript{11,26,28} However, polysomnography studies of infantile RMD are too few to prove that rhythmic movements are exclusively related to the superficial sleep.\textsuperscript{28}

**Table 2—Episodes of Rhythmic Movements in Sleep Stages**

<table>
<thead>
<tr>
<th>Pt</th>
<th>NREM</th>
<th>NREM</th>
<th>NREM</th>
<th>REM</th>
<th>W</th>
<th>Total</th>
<th>Duration, min:sec</th>
</tr>
</thead>
<tbody>
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<td>1</td>
<td>0</td>
<td>65</td>
<td>43</td>
<td>4</td>
<td>4</td>
<td>116</td>
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<tr>
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<td>16</td>
<td>14</td>
<td>4</td>
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<td>9</td>
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</tr>
<tr>
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<td>4</td>
<td>8</td>
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</tr>
<tr>
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<td>6</td>
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<td>0</td>
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<td>0</td>
<td>18</td>
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<tr>
<td>5</td>
<td>5</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
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<td>2:30</td>
</tr>
<tr>
<td>8</td>
<td>25</td>
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<td>3</td>
<td>20</td>
<td>2</td>
<td>71</td>
<td>85:00</td>
</tr>
<tr>
<td>9</td>
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<td>0</td>
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<td>0:06</td>
</tr>
<tr>
<td>10</td>
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<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
<td>0:17</td>
</tr>
</tbody>
</table>

Pt refers to patient number; W, wakefulness; NREM, non-rapid eye movement sleep; REM, rapid eye movement sleep.

**Figure 2—Rhythmic movements in rapid eye movement sleep.**

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<table>
<thead>
<tr>
<th>Table 3 — Sleep Parameters</th>
<th>Patients</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>1</td>
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<tr>
<td>Total sleep time, h:min</td>
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<tr>
<td>Sleep-onset latency, min</td>
<td>4.5</td>
</tr>
<tr>
<td>Sleep efficiency, %</td>
<td>97</td>
</tr>
<tr>
<td>Sleep stage, % of total sleep time</td>
<td></td>
</tr>
<tr>
<td>Wake</td>
<td>1.6</td>
</tr>
<tr>
<td>NREM 1</td>
<td>0.3</td>
</tr>
<tr>
<td>NREM 2</td>
<td>24.0</td>
</tr>
<tr>
<td>NREM 3</td>
<td>22.9</td>
</tr>
<tr>
<td>NREM 4</td>
<td>31.4</td>
</tr>
<tr>
<td>REM</td>
<td>19.6</td>
</tr>
<tr>
<td>REM latency, min</td>
<td>97</td>
</tr>
<tr>
<td>Movement time, %</td>
<td>0.1</td>
</tr>
</tbody>
</table>

W refers to wakefulness; NREM, non-rapid eye movement sleep; REM, rapid eye movement sleep.

The diagnosis of RMD seldom poses a problem because of the stereotyped nature of the movements. RMD may be confused in infants with bruxism or thumb sucking, and, rarely, periodic leg movement disorder may produce similar features.² ³

As for epileptic disorders, infantile spasms and nocturnal frontal lobe epilepsy might resemble rhythmic movements.² For this reason, we used more leads to monitor EEG activity. We found epileptic discharges during NREM stage 2 sleep in 2 patients without any connection with rhythmic movements. Rhythmic-movement episodes were never preceded by autonomic function or EEG changes, as nocturnal seizures characteristically are.² ³² ³³

The age of onset was also atypical for nocturnal frontal lobe epilepsy and so was the occurrence of rhythmic movements in REM sleep in some patients.

However, some authors believe that RMD has such typical anamnestic and clinical data that no real differential diagnosis is needed there.¹³

Basically, as to the duration of movements, we observed 2 types of rhythmic movements: longer rhythmic movements occurred in wakefulness or NREM stage 1 sleep, while shorter rhythmic movements occurred during NREM stage 2, NREM stage 3/4, and REM sleep. Although the shorter episodes were
not usually preceded or followed by arousals, we observed that some rhythmic movements were preceded by a very short period (1-2 seconds) of faster EEG activity that could not be rated as an arousal according to standard scoring rules defined by American Sleep Disorders Association.

Our observation of short periods of electromyogram activation in REM sleep, whether or not connected with rhythmic movements (as found in 4 cases), is in contrast with other reported cases of rhythmic movements during REM sleep, in which no muscle-atonia changes were noted.11,23,24,28 Nevertheless, some authors speculate that cases with rhythmic movements predominating in REM sleep may be a variant of REM sleep behavior disorder.11

According to some studies, the prevailing type of rhythmic movements varies with age. Bodyrocking is the most frequent before 12 months of age. It is then followed by headbanging, and, at the age of 2 to 3 years, by headrolling.3 In our series, headbanging was as frequent as bodyrolling. During REM sleep, rolling movements alone were observed. Similar findings have been reported by some authors,11,26 although others have also found headbanging and bodyrocking in REM sleep.14,27,28

The pathophysiologic mechanisms of rhythmic movements remain obscure. The hypothesis of the origin of rhythmic movements of infancy includes learned “self-stimulating” or “auto-erotic” behavior connected with pleasurable feelings in what is thought to be a tension-releasing maneuver of a high hypnogenic potential.10,11 The maturational role of rhythmic movements in early psychomotor development by means of vestibular apparatus stimulation10,11 is also hypothesized, although this only applies to bodyrockers.12

However, these hypotheses do not explain how these voluntary or at least partly voluntary movements become integrated in the sleep process as involuntary movements not preceded or followed by arousals as defined by American Sleep Disorders Association. This is a feature similar to tic movements, which are also incorporated into all sleep stages—especially REM sleep—yet do not lead to arousals. The described significant drive and urge to perform rhythmic movements also resembles tics except that there are no rhythmic movements in the latter. The pathophysiologic mechanism of tics can be traced to decreased motor-inhibition control.34-36

This lack of the correlation between American Sleep Disorders Association-defined arousals and tic movements supports the idea of the subcortical part of corticostriato-thalamocortical circuits as having a role to play here. A similar motor-pathway involvement may conceivably account for rhythmic movements. The role of basal ganglia in rhythmic movements has been considered.3,5,7 Speculating on the subject, Kohyama et al28 examined the frequency of gross movements during sleep in 2 patients with RMD but failed to find any changes. Obviously, more studies are necessary.

However, a recent case report has demonstrated a close relationship between phases A2 and A3 of the cycling alternating pattern of rhythm movements during NREM sleep.38 The result was obtained with a different kind of analysis, we probably could have revealed more clearly the relationship between rhythmic movements and cortical or subcortical arousals.

Another moot question concerns factors causing the persistence of rhythmic movements beyond infancy. Familial forms have been described at a rate of up to 20%.6,16-20 We also found positive family history in 2 patients. Perinatal history was positive in 5 patients, raising the suspicion of possible functional brain damage sustained during this period.

RMD in its typical form is considered to be a benign and self-limited disorder marked by no daytime symptoms, except in cases of violent rhythmic movements, which can lead to head and eye injuries. The results of our investigation suggest a different idea. All patients had some daytime complaints: attention difficulties, hyperactivity, neurotic symptoms, morning headache, fatigue, and sleepiness. A similar observation has been made by other authors who have reported a number of problems, mostly in adult patients with rhythmic movements: anxiety, depression, daytime sleepiness, irritability, fatigue, poor concentration, and poor sleep quality.11,21,22,26,38 Hence, it is doubtful that persistent RMD is really a benign condition; instead it may also have some health consequences. Thus, careful evaluation of daytime symptoms, especially in children, and detailed psychologic investigation seem to be very important. Whether these symptoms are connected to insufficient sleep quality caused by rhythmic movements or constitute an independent feature remains unclear. More reports are necessary for evaluation.

In the age group of 19 to 24 years, we found neurotic symptoms, depression, obsessive-compulsive disorder, while 6 out of 7 school children had ADHD, predominantly of the inattentive type. The occurrence of ADHD in children with RMD has also been previously reported in 3 patients.23

Although a relationship between periodic limb movement disorder and ADHD has been reported,24-41 the common pathogenetic mechanism is not yet clearly elucidated. We can hypothesize that functional impairment or immaturity of premotor and striatal circuits may be involved in RMD, as well as periodic limb movement disorder and ADHD, or that rhythmic movements might be another type of sleep disorder similar to sleep-disordered breathing, snoring, or periodic limb movement disorder causing ADHD-like symptoms (secondary ADHD).42,43

Concurrently with recent reports, the need arises to revise the definition of this type of parasomnia and to continue research into its pathogenetic mechanisms and possible subtypes in relation to sleep stages and conceivable health risks.

REFERENCES