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NON-EPILEPTIC PAROXYSMAL EVENTS IN CHILDREN: STRUCTURE AND PHENOMENOLOGY

II. RHYTHMIC MOVEMENT DISORDER

Abstract. Parents, pedagogues and doctors often face ambivalent conditions of children difficult to be diagnosed as normal, adaptive or pathological. Non-Epileptic Paroxysmal Events (NEPE) occupy a special place among them. The given research focuses on the description of the main non-epileptic paroxysmal events in children which are often observed in everyday professional activity of pedagogues, psychologists and pediatricians and may be difficult to interpret. The authors have undertaken an analysis of diagnosability of NEPE at the specialized neurological department of the city children's hospital in 2016. Four out of 78 children with the admission diagnosis of NEPE arrived from children's preschool institutions where their paroxysmal disorders caused special anxiety and worry. The NEPE was diagnosed in 53,8 % of cases; in 46,2 % of cases the disorders failed to be differentiated. The study revealed 8 children with nocturnal NEPE (10.3%; 6 of them – with benign alternating nocturnal hemiplegia, benign sleep myoclonus, masturbation, sleep apnea syndrome; 2 children – with startle and dyspnea with eye adversion). The given article deals with the paroxysmal events observed during sleep which occupy a significant portion of time in the structure of the cycle "sleep – waking up – wakefulness" in infants (up to 2/3 of the day). Alongside movements which are typical of children at various phases of sleep, a number of infants demonstrate paroxysmal events with controversial interpretations. Our research reveals their structure and manifestations. In the overwhelming majority of cases, the prognoses for these conditions are favorable if they are treated adequately by parents, pedagogues and doctors.

Keywords: non-epileptic paroxysmal events; non-epileptic paroxysmal disorders; pediatrics, dreams; motor acts.

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People spend a significant part of their lives sleeping. There are controversial opinions about the age-related norms of duration of sleep every night; sometimes they are unduly prescriptive. Anyway, it seems that we spend about one third of our life asleep. But 20 years of teaching neurology and children's neurology to more than 5,000 post-graduate students-neurologists, pediatricians and neonatologists have shown that none of them studied physiology and pathology of sleep at medical higher education institutions.

E. Bathory et al. [7] argues that during regular examination of children, pediatricians often give inadequate assessment of sleep and its disorders. According to the authors, this is due to the fact that the problems of sleep are paid little attention to at medical universities and during residency.

Table 1. Equivalents of the Russian word "*son*" in different languages

Language	"*Son*" as a process	"*Son*" as a dream
English	sleep, slumber	dream, vision
German	Schlaf	Traum, Traumscheinung
French	sommeil	rêve, songe, on(e)irisme

While looking at the development of the notions about sleep, it is necessary to note the semantic peculiarity of the Russian lexeme "*son*". In Russian, the word "*son*" means both the state of sleeping and a dream. In spite of the fact that the word "*snovidenie*" exists in Russian alongside the word "*son*", it is rarely used in everyday speech and has a somewhat elevated meaning. When we have had a dream, we usually say "*mne snilsya son*". Meanwhile, in many languages "*son*" as a process and "*son*" as a dream are different words (see Table 1).

The given paper deals with sleep mostly as a process or state. There are several definitions of sleep, but the one given below is more widely accepted.

Sleep is a natural, regular periodic state of rest of the body and mind, during which the eyes, as a rule, are closed, consciousness and arbitrary movements are either absent or insignificant, and passing dreams appear from time to time [16].

The question about the time of emergence of sleep as a process or state still remains open. According to the scheme of emergence of the basic neurological functions in the fetus, the most important time of sleep formation begins at about the 20^{th} week of gestation, when rapid eye movements appear, and the 34^{th} week of gestation which marks the period of primary sleep stage (or phase) distinction [12; 17].

Stage 1 is the transition from wakefulness to the onset of sleep. It is characterized by irregular low voltage activity on the electroencephalogram (EEG). Stage 2 is reflected in light, *superficial sleep* showing bursts of 12-14 Hz sinusoidal waves called sleep spindles and high-voltage biphasic waves called K complexes in the EEG. At Stage 3, there are sleep spindles and high-amplitude, slow delta waves in the EEG. At Stage 4, corresponding to deep sleep, slow-wave activity increases and dominates the EEG record. Stages 3 and 4 in humans are sometimes called slow-wave, deep, quiet, synchronized, orthodoxical sleep. Finally, a special fast, active, desynchronized, paradoxical stage of sleep characterized by irregular low voltage EEG activity and rapid movements of the eyes (REM stage) is distinguished.

Different points of view exist about the daily norms of sleep duration. Due to the dependence of the character and quantitative sleep parameters on a number of factors, these norms are always correlated with the contingent under study. In Figure 1, we can see the data obtained for Australian children.

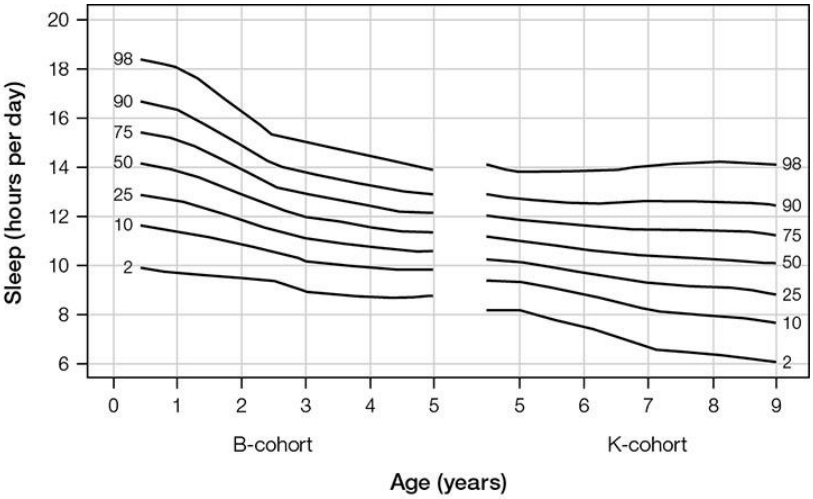

Figure 1. Age-related dynamics of the daily sleep duration for two age groups (cohorts) of Australian children [16].

According to R.H. Adair et al. [4], the healthy full-term newborn infant sleeps from 16 to 18 hours per day in several separate periods throughout the day. The pattern "sleep – wakefulness" is irregular, the longest sleep period is 2.5-4 hours and is connected with feeding. Approximately up to three years of age, the child needs more hours of sleep than wakefulness each day [10].

A. Rechtschaffen et al. [21] singles out the following functions of sleep: conservation of energy, cognition, thermoregulation, neural maturation and mental health. According to the conception of O. Pompeiano, [20], in the course of evolution, sleep emerged with the purpose of limiting motor activity as a kind of animal's rest from movement. Due to the scarcity of behavioral opportunities of the archaic animal which were actually limited to the dichotomy mobility/immobility, the development of sleep facilitated protection of the primitive organism from physical fatigue. Nevertheless, normal sleep is accompanied by motor activity.

A. Z. Golbin [2] refers general movements of body and limbs without changing posture, relatively isolated movements (with the head or limbs), local separate movements (facial expressions, finger and toe movements), separate paroxysmal movements (startle, jitteriness), rhythmic movements (sucking) and isometric muscle tension to com-

paratively simple *motor responses during sleep*.

Covering oneself with a blanket, manipulations with clothes, stretching, and movements aimed at taking a comfortable posture may be referred to motor acts with elements of adaptive behavior.

Somatic-vegetative responses include panting, snoring, noisy exhalations and inhalations, respiratory arrhythmia, coughing, swallowing, hiccupping, activization of intestinal peristalsis, erection, etc.

The phenomena accompanied by activation of the vocal-motor apparatus include moaning, inarticulate muttering, and articulate speech.

The prevailing distribution of various motor phenomena across sleep phases according to I. A. Vakhrameeva [1] is shown in Figure 2.

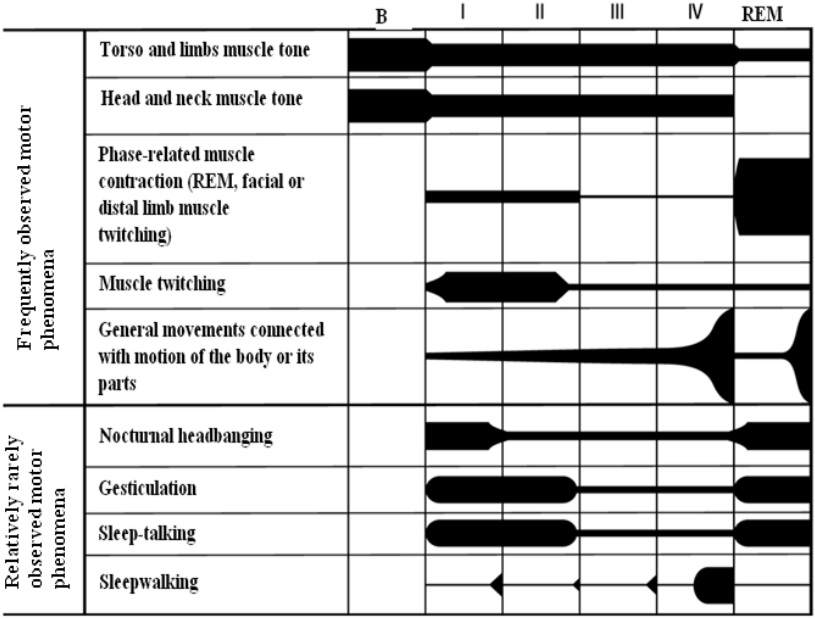

Figure 2. Distribution of motor phenomena across sleep phases (according to I. A. Vakhrameeva [1]).

Note: B — wakefulness

Thus, even in case of typical development, we can observe a significant variety of movements during sleep. Separate motor phenomena have a sub-optimal nature and may be treated as personal specific traits, but given they are stable, total and leading to marked disorders of the cycle *sleep – waking up – wakefulness*, they obtain abnormal and pathological significance.

One third of the population of the planet have problems with sleep brought about by social, economic and medical causes [8].

According to various sources, 20 through 50% of children have sleep disorders [7; 9; 13; 18; 19; 23].

Widespread incidence of sleep disorders in children and numerous ambivalently interpreted cases of sleep impairment have determined the object of the given research.

The aim of our research is to describe non-epileptic paroxysmal events (NEPE) observed in children during sleep which are difficult to interpret by the parents and medical and pedagogical specialists (especially in pre-school and social care institutions).

Over the period from January 1 to December 31, 2016, 78 children up to 4 years of age were admitted to the neurological department of the City Children's Hospital of Saint Olga (Saint Petersburg) with paroxysmal mental disorders. Four of them arrived from children's preschool institutions accompanied by medical personnel or parents in connection with emergence of paroxysmal disorders at crèche or children's home.

The general information about the children under examination is presented in Table 2.

The scope and the nature of examination were described in our previous article [4].

Table 2. Characteristics of children under examination

Parameter		M ($X_{min.}$ — $X_{max.}$)
Sex	boys	39
	girls	39
Gestational age, months		35,5 (29—42)
Postnatal age, months		25,5 (1—50)
Optimality of the course of pregnancy, %		83,5 (70—97)
Optimality of the course of birth, %		80,5 (61—100)
The *Apgar score* 1'		5 (1—9)
..... 5'		6,5 (4—9)

The research results showed that 22 children (28.9 %) among those with paroxysmal disorders typified as NEPE did not have neurological deviations; the other infants demonstrated various causal deviations of the neurological status; 12 children (15.9 %) had multiple deviations. Ultrasonographic examination showed normal brain structure in the majority of children (55.3 %); mild brain ventricular expansion was predominant among the deviations observed (22.4 %).

EEG corresponded to the age-related norm in 73 children (96%); three children (4%) showed delay of bioelectrical activity formation, which served as an additional argument in favor of referring the paroxysms to NEPE.

The structure of the diagnosed NEPE is presented in Table 3.

Таблица 3. Character of paroxysmal consciousness and movement disorders in children under observation

Character of paroxysmal disorders	Number	Percentage, %
NEPE differentiated into:	42	53.8
– breath-holding spells	9	11.5
– benign paroxysmal eye phenomena	6	7.7
– Fejerman syndrome	11	14.1
– benign paroxysmal torticollis (retrocollis)	4	5.1
– *benign* nocturnal *alternating hemiplegia*	2	2.6
– mild hyperekplexia	2	2.6
– masturbation	2	2.6
– benign *sleep myoclonus*	1	1.3
– spasmus nutans	1	1.3
– sleep apnea	1	1.3
– paroxysmal *dystonia attacks*	1	1.3
– jitteriness	1	1.3
– startle response	1	1.3
Undifferentiated NEPE	36	46.2

The present research revealed 8 children (10.3%) with nocturnal NEPE (6 of them were differentiated into: *benign* nocturnal *alternating hemiplegia*, benign *sleep myoclonus*, masturbation, sleep apnea; 2 of them were not differentiated: with startle and dyspnea with eye adversion).

Nocturnal NEPE are specifically systematized and interpreted.

A. Z. Golbin [2] considered them as stereotypies during sleep and singled out rocking, banging, folding, shuttling, sucking fingers at night, masturbation, etc.

Further development of the theory of NEPE allowed the researchers to single out periodical and rhythmic parasomnias and evolutional episodic paroxysmal sleep phenomena [14; 15].

In correspondence with the ICSD-3 [6], these phenomena can be distributed between parasomnias and sleep-related rhythmic movement disorders (RMD).

RMD constitute a group of stereotypical repeated movements involving long muscles such as the head and neck muscles, etc. They include rocking, banging, shuttling, folding, etc. Historically, these movements are called *jactatio nocturna* (from Latin *jactare* — rhythmic rocking).

Some forms of rhythmic activity can be observed in 2/3 of infants up to 9 months of age. Further on, the frequency of manifestation of these events decreases: by the age of 18 months they are demonstrated by less than 50% of toddlers, and by the age of 4 years – less than 8% of children [11]. It was believed over several decades that RMD were exclusively typical of children with intellectual disability or emotional deprivation. The given disorders are revealed more often in orphans. Children with emotional deprivation or brain lesions are characterized not only by stereotypical self-calming and self-stimulating behavior but also by self-harming activity which is practiced during wakefulness as well. However, this point of view is no longer domineering [14].

Rocking consists of rhythmic pendulous head or body movements of different amplitude from side to side with the frequency of 0.5-2 Hz. The length of rocking movements depends on the disorder severity (from several minutes to several hours); rocking becomes asymmetric as the condition grows more severe. As a rule, rocking is observed until the age of 1 year (sometimes *in utero*), demonstrates the peak of manifestation at the age of 6 months, and may disappear at any age. Rocking usually starts in the drowsy state and vanishes during NREM Stage 4. The transitions between sleep phases are more favorable for rocking.

We have not discovered any connection between rocking and certain medical or social factors. Still there are data about possible inherited susceptibility to rocking and about its emergence (as well as disappearance) after an event significant for the child (illness, parting with parents, moving to another location, etc.).

Mild rocking can be described as rhythmic pendulous head or body

movements of different amplitude from side to side with the frequency of 0.5-2 Hz. The movements are smooth, stereotyped and rhythmic, one series of movements lasts 5 through 10 minutes. The nature of rocking movements is individual and stable; the beginning and the end of each episode are soft. The limbs and the torso are either immobile, or slightly move to the rhythm of the head movements. Rocking is observed at the moments of falling asleep or arousal from sleep, and is not typical of wakefulness. Older children and adults feel pleasure and comfort from rocking when they wake up, and they reproduce rocking readily. During wakefulness, such children are active enough.

Moderate rocking involves the limbs and the torso into the action. Torso rockings are sharp and follow the movements of the head. The arms also reproduce the rhythm of the head rocking; the elbows are bent. The episodes become more intensive and form "clusters". The length and intensity of the episodes increases during the night. Rocking passes on to the period of wakefulness when the child is in the state of excitation. The infant no longer relaxes; moreover, he is tense during rocking. The involvement of the upper limbs into the action, and movement asymmetry are typical of mild rocking. The length of each cluster reaches half-hour, and the frequency approaches 60-90 movements per minute. The clusters begin and end sharply, as if they were "switched on" and "switched out". The child's behavior during wakefulness remains normal.

Severe rocking demonstrates movements so intensive that they might be taken for a paroxysm. We observe sharp movements of the head; the arms and the torso are thrown apart. The arms may be spread apart with clenched fists, or may be bent at the elbows and held close to the torso. In this rocking phase, the movements are predominantly asymmetric (and the asymmetry does not depend on the hemispherical dominance of the child). The length of a rocking episode may be several hours and may include up to 2,000 movements without stopping. The EEG changes of the children with severe rocking (dysrhythmia, decrease of the wave amplitude) are recorded, as a rule, only in severe forms of stereotypies. During pauses between strong rocking movements, EEG registers transitory patterns of Stages 2 and 3 of NREM sleep. Severe rocking can cause giddiness and nausea. During wakefulness, the child may experience problems with learning, attention deficit, and drowsiness; later on, the intellectual abilities may develop normally, but there might be problems with behavior [3].

Banging are stereotypies when the child, lying in the prone posi-

tion, bangs his head on the pillow propping himself up on stretched arms. In cases of mild manifestation, and in older children, banging involves only movements of the head. In severe cases, on the contrary, the stereotypy engages the whole upper part of the torso. The movements may increase and decrease in amplitude and frequency. The forehead, cheeks or temple may touch the pillow. At first, banging lasts 5 through 10 minutes continuously, with the frequency of 60-65 movements per minute. Then the frequency increases up to 80-89 movements per minute, forming clusters. Such clusters make up episodes lasting from 1 minute to 2 hours. Night episodes may continue till late in the morning and appear in sleep or drowsy state. Attempts to calm the baby from the outside may stop the movements for some time, but they are soon resumed with increased intensity. Banging ceases as soon as the baby is laid in the supine position. When the infant turns to lie on the stomach, he resumes the prone position and starts rolling his head energetically and asymmetrically back and forth until the head bangs on the pillow. In most cases, banging begins in infants at the age of about 1 year with a period of warning signs; these stereotypies are more saliently connected with organic disorders of the nervous system (specifically, with perinatal ones), and may be attributed to the dyssynchrony of development or lesions of the vestibular system or the cerebellum. EEG detects paroxysmal high amplitude slow wave activity and sharp waves. We have noted instability of Stage 3 of NREM sleep.

The *shuttling phenomenon* consists in the infant's rocking back and forth on hands and knees. In some cases these movements are interpreted by the surrounding people as "masturbation", which is disproved by sleep video monitoring. The movements forward are, as rule, faster than the movements backward. The head is against a wall or pillow. The child turns the head with the pressure on the head growing. The children do not get pleasure from these movements, remember them vaguely, but try to get rid of the accompanying giddiness, anxiety and unpleasant tingling. These movements appear in children aged 1.5-3 years without any evident cause, sometimes after certain somatic diseases. It is supposed that the factor of inheritance may be associated with the development of these stereotypies. The EEG of the children suffering from the given stereotypies, as a rule, corresponds to the age-related norm. The sleep structure shows an increase of the length of drowsing and a shorter phase of REM sleep.

The phenomenon of folding presupposes rhythmic raising the torso and knees simultaneously from the

supine or sitting position, with a tendency to increase the amplitude and frequency of movements. Typically these movements will occur just before sleep begins during Stage 1 and vanish at transition to Stage 4. It is a rare, original stereotypical sleep disorder described by A. Z. Golbin [2] in children suffering from somatic (as a rule, allergic) diseases, hyperactivity, emotional lability and having complicated perinatal anamnesis with phenomena of inhibition and muscular hypotonia during the first months of life.

Among other rhythmic movement phenomena A. Z. Golbin [15; 16] distinguishes arms and legs swinging, hitting movements, excessive sucking of thumb and tongue, chewing, vocalization, hair pulling, etc.

The EEG of rhythmic motor phenomena shows patterns associated with impairment of maturation of bioelectrical activity of the brain, predominantly light and diffuse. These deviations are more often observed in cases of rocking. They demonstrate epileptic activity very rarely.

On the whole, the analysis of development of the persons with *jactatio nocturna* testifies to the fact that they are lively and active people. In their everyday life, they are engaged in kinds of activity associated with rhythm (dances, music, especially jazz). Such children, adolescents and adults sometimes have rare problems of impaired nasal breathing and otitis; their clinical picture includes soft neurological signs, specificity of interhemispheric relations, perception disorders, anxiety, etc. [14].

Thus, rhythmic movements associated with sleep are frequent children's conditions worrying close adults, caregivers and doctors and sometimes leading to unnecessary hospitalization and inadequate intervention. These non-epileptic paroxysmal events need delicate treatment and have absolutely favorable prognosis.

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