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Non-epileptic paroxysmal events in early childhood and role of EEG: A single center experience

Erken çocukluk çağında epileptik olmayan paroksismal olaylar ve EEG'nin rolü: Tek merkez deneyimi

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Abstract

Aim: Non-epileptic paroxysmal events are the most frequently movement disorders mimicking epilepsy. Our aim in this article is to increase awareness among physicians by sharing our clinical experience; to reduce unnecessary anti-epileptic use and the number of examinations.

Methods: In total 73 patients were included in the study. Patients were evaluated according to detailed anamnesis, video recordings, laboratory findings and electroencephalography (EEG) findings.

Results: The most common diagnosis in patients involved in the study was breath holding spells. The youngest age group was benign sleeping myoclonus. None of the patients had epileptiform activity in the EEG.

Conclusion: Non-epileptic paroxysmal events commonly involve the unnecessary use of anti-epileptic medications due to mimicking of epilepsy. This study has shown that detailed anamnesis, EEG findings, and increasinguse of mobile phone video reduce unnecessary treatment and examination in these patients.

Keywords: Paroxysmal, electroencephalography, children

Öz

Amaç: Epileptik olmayan paroksismal olaylar, epilepsi ile en sık karıştırılan hareket bozukluklarıdır. Bu yazıdaki amacımız klinik deneyimimizi paylaşarak hekimler arasında farkındalığı arttırmak; gereksiz anti-epilepik kullanımını ve tetkik sayısını azaltmaktır.

Yöntemler: Çalışmaya 73 hasta dahil edildi. Hastalar ayrıntılı anamnez, video kayıtları, laboratuvar bulguları ve elektroensefalografi (EEG) bulgularına göre tanı açısından değerlendirildi.

Bulgular: Çalışmaya dahil edilen hastalarda en sıktanı katılma nöbetiydi. En küçük yaş grubu benign uyku myoklonisiydi.Hastaların hiçbirinin EEG'sinde epileptiform aktivite saptanmadı.

Sonuç: Epileptik olmayan paroksismal olaylar, epilepsi ile sık karıştıklarından sıklıkla gereksiz anti-epileptik tedavi kullanımı söz konusu olmaktadır. Bu çalışma, ayrıntılı anamnez, EEG bulguları ve giderek yaygınlaşan video kullanımının böylesi hastalarda gereksiz tedavi ve tetkik gereksinimini azalttığını göstermiştir.

Anahtar Kelimeler: Paroksismal, elektroensefalografi, çocuk

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Introduction

Non-epileptic paroxysmal events (NEPE) in early childhood as a complicated case involving the intermittent repetitive motor movements, behavioral changes and somatic symptoms cause disquiet among parents and physicians in most of the time [1]. Symptoms diminish with age, even disappear before the school ages, and often do not require treatment which is the most important characteristic in most of the cases. Additionally, some patients are subjected to treatment due to misdiagnosis of epilepsy. For differentiating these symptoms from epileptic seizures, the absence of electroencephalographic (EEG) epileptiform changes in the event of an epileptic seizure proves itself useful in diagnosis. The most crucial step for diagnosis is the detailed anamnesis. Frequent NEPE in early childhood have a wide spectrum and are composed of benign sleeping myoclonus (BSM), benign paroxysmal vertigo (BPV), breath-holding spell (BHS), night terrors (pavornocturnus) and infantile masturbation [2, 3]. Less commonly observed symptoms include chills, hyperekplexia and paroxysmal tonic upgaze. Nowadays, with the widespread use of camera devices such as mobile phones, the correct diagnosis of such events can be established easily [4].

In this particular study, we aimed to raise awareness of pediatric physicians by showing the clinical characteristics, EEG findings and established diagnoses of the patients who applied to pediatric neurology clinics; thus, making contributions to reduction of misdiagnosis, unnecessary examination, excessive costs of treatment.

Material and Methods

In our study, the files of the patients who had BSM, BPV, BHS, pavornocturnus and infantile masturbation were reviewed retrospectively in Kahramanmaraş Sütçü İmam University Medical School Pediatric Neurology Clinic between June 2017 and April 2018. The study was performed according to the declaration of Helsinki. Written consent was taken from parents of the patients.

The study included patients admitted our pediatric neurology clinic, normal neuromotor development, at least one family member or guardian who was able to identify habitual events. Patients' diagnoses consisted of BHS (cyanotic type-pallid type), BSM, pavornocturnus, BPV and infantile masturbation. Patients with pathologic neurological examination, patients with neuromotor growth retardation and patients whose file information was not available were excluded from the study.

We retrospectively reviewed the medical records of a total of 87 cases. Fourteen patients were excluded from the study; so, a total of 73 patients were included in the study.

The detailed anamnesis of patients, duration of complaints, EEG records, laboratory findings, physical examination findings and images obtained by families and/or findings acquired during EEG were recorded. Two types of BHS are present based on the color of the child during the apneic episode following the end of prolonged expiration either pale (pallid attacks) or blue (cyanotic attacks) [4]. The 3rd Beta criteria determined by the International Headache Society for BPV were taken into consideration [5]. Patients with low hemoglobin concentration and low ferritin levels than (<10 ng/dL) were evaluated for iron deficiency anemia. Dallman anemia criteria for hemoglobin concentration were taken as reference according to age (6 months-4.9 years= 11 g/dL, 5-11.9 years= 11.5 g/dL) [6].

Statistical Analysis

Data analysis was done by the SPSS 22.0 package program. Normally-distributed continuous variables were expressed as mean ± standard deviation. Continuous variables without normal distribution and categorical variables were expressed as median and frequencies with percentages, respectively.

Results

There were a total of 73 patients included in the study. The average age of these patients was 14.42 ± 10.24 months (45 days-49 months). Thirty nine (53.4%) of the patients were male and 34 (46.5%) were female. Female to male ratio was higher in patients diagnosed with infantile masturbation (F / M: 2/1) contrary to the other patients in which higher male to female ratios were present.

The most commonly established diagnosis was BHS (n= 51) followed by pavornocturnus (n= 9). The patients with BSM constituted the youngest age group (mean= 53.2 ± 12.8days) and the infantile masturbation group had the oldest age group (mean= 41± 8.6 months) (Table).

All of the patients' EEG records were examined. Non-epileptic paroxysmal activity was detected in EEG in 8 patients (10.9%). None of the patients showed any epileptic abnormalities on EEG.

Table: Demographic and clinical characteristics of the patients due to non-epileptic paroxysmal events.

Diagnosis		n (%)	Male sex (%)	Age (M/D) β	Attack frequency (D/M/Y)	Duration of Symptoms (min)	Sleep-awake
BHS		51 (69.8)	52.9	19.5±11.6 M	7.6±4.3 M	3±4.6	Awake
	Cyanotic	32 (43.8)	53.1	20.2±7.4 M	8.1±5.2 M	3±4.3	Awake
	Pallid	19 (26)	52.6	17.4±8.3 M	7±3.4 M	3±5.2	Awake
Pavor nocturnus		9 (12.3)	55.5	38 ±10.7 M	8.3±6.3 M	10±8.3	Sleep
BSM		7 (9.5)	57.1	52.3±12.8 D	4±4.8 D	1±2.3	Sleep
BPV		3 (4.1)	66.6	22.3±9.3 M	4.2±3.5 Y	5±4.1	Awake
IM		3 (4.1)	33.3	41±8.6 M	5.2±8.2 D	5±3.8	Awake

β: mean±standard deviation, BHS: Breath-holding spell, BSM: Benign sleep myoclonus, BPV: Benign paroxysmal vertigo, IM: Infantile masturbation, D: day, M: month, Y: year, min: minutes

Neuroimaging was performed on 3 patients with BPV diagnosis. Pathology was not observed in imaging findings. Iron-replacement therapy was administered to 43 patients who applied for breath-holding spell and to those who had iron deficiency anemia. Eight patients without anemia were recommended and treated with piracetam (Nootropil®, 5-10 mg/kg/dose, per oral, three times a day, UCB Pharma, Istanbul, Turkey). A significant decrease in complaints was observed in 3-month follow-ups of the patients.

In pavornocturnus, the attack interval was one time during the day. Families were given recommendations. All of the patients had regression in their symptoms after 3 months. Complaints of patients diagnosed with BSM ended before the patients were 3 months old. Cyproheptadine (Sipraktin®, 2-4 mg/dose, per oral, twice a day, I.E. Ulagay, Istanbul, Turkey) therapy was initiated for all BPV patients. At the end of the third month of the test, there was a decrease in the frequency of attacks. All patients with infantile masturbation had diaper dermatitis and patients' families were given recommendations. There was a decrease almost %90 in the interval of attacks during the 3-month follow-up period in BSM patients. Also, the number of attacks in patients with BSM and infantile masturbation was significantly higher than the other patients.

Discussion

Non-epileptic paroxysmal events include an episodic phenomenon, a motor phenomenon with a variable duration and a generally stereotypical character. The diagnosis is based on detailed anamnesis and attentive observation. Clinical spectra are quite extensive. Frequencies reported in various studies that were carried out by Kotagal et al. [7] was at 15% while the study carried out by Patel et al. [8] demonstrated as 3.5%. Duration, location, shape, time of occurrence, state of consciousness of the attacks; between the epilepsy and the NEPE diagnosis, may cause hesitance among physicians. In this case, differential diagnosis can be established by recording the seizure through a video recording device and evaluating the EEG at the same time [1, 2].

BHS is a paroxysmal disorder that is triggered by emotional and / or physical stimuli that occurs approximately at 4.6% during early childhood. It is more commonly observed among males. The reported age range varies between the 3rd month and up to 6 years, but in most of the cases range is between 6 months and 36 months [4, 9]. The majority of patients presenting with BHS have a seizure of cyanotic type followed by pallid type seizures. BHS begins with a warning period, followed by crying. The triggering factors during the stimulation differ according to the types of seizures. BHS, a cyanotic type, is a psychogenic stimulus that induces seizure, whereas the pallid type of trauma is anterior [10, 11]. In our study, the number of male patients was higher and cyanotic type seizures were detected in the majority of patients in accordance with the literature. The vast majority of patients were composed of individuals younger than 3 years of age. No epileptic abnormality was observed during any EEG recording. In particular, most patients benefit from iron therapy.

Pavornocturnus is a sleep disorder that occurs in the non-REM sleep period, followed by screaming, cold sweating, crying and hallucinations about 1.5-2 hours into sleeping. It is most commonly seen between 4-6 years of age and in males. It is typically observed once in the night. As it may be limited to a few minutes, the time may be longer than expected. Patients may not recognize their family during the time of the attack and do not remember any of them the next day [12]. EEG recordings of all patients in our study were normal. Suggestions such as reducing the duration for watching television, and not watching videos from mobile phones were made. There was a decrease in the number of attacks in the follow-up time.

The International Headache Society described BPV as a group of periodic syndromes in childhood and stated that it was a leading symptom of migraine [5]. BPV begins during the infancy period; as episodes of sudden onset of dizziness and not exceeding a few minutes. Attacks usually begin to be observed at 1-2 years after walking and may last up to six years. Nausea, vomiting, paleness is usually evident. During events the child is awake, exhibits panic-like behavior and suddenly scared and refuses to move until the event is over. If the child is tried to be carried during this period, unbalanced movement and behavior is usually observed. Electroencephalography and magnetic resonance imaging are normal. Cyproheptadine and diphenhydramine treatment were reported to be beneficial [13]. In our study, magnetic resonance imaging and EEG findings of three patients were normal and patients benefited from cyproheptadine treatment.

Infantile masturbation is defined as the child's self-stimulating pleasure behaviors. The age of onset is between 3 months and 5 years and it is observed more frequently in females. When the patient is sitting or lying down, stretches legs, compresses breath, blushing occurs. The event lasts a few minutes and can be interrupted. It can be repeated 15-20 times a day. When infections of the urinary tract, such as diaper dermatitis, cause itching, the child accidentally discovers that he or she is pleased with the movements he or she makes in order to get rid of the irritation. With behavioral therapy, this can be eliminated [14, 15]. The number of female patients in our study was high and neurological examinations and EEG recordings were normal. Diaper dermatitis was mentioned in 3 of the patients. The parents were warned and informed about diaper dermatitis and urinary tract infection. A decrease in the number of complaints in the control was detected.

BSM is a myoclonic beat during the newborn and premature infantile period, especially when sleeping, repetitive, high frequency, for seconds or for minutes. It is thought that the neuronal structure that provides motor control during sleep is not mature and is caused by genetic factors. When the child wakes up, episode ends. The most important characteristic is not seen outside sleep. These attacks usually disappear when the child is 4-6 months old. Treatment is unnecessary [16]. Patients involved in our study constituted the youngest age group of patients. Neuromotor development of the patients was evaluated as normal. EEG examinations showed no epileptiform abnormality with movement. In patients with tumors, neuromotor development was consistent with months, and when they were 3 months old, movements disappeared.As a result, during early childhood it is disquieting for both parents and physicians because it can get involved with NEPE, especially epilepsy. With the increasing EEG requirements in recent years and the increasing use of mobile camera phones, great convenience has been provided to these patients. Accurate diagnosis and treatment costs are at the least. Misdiagnosis can cause the child to change his or her life style or future plans. This is an intense source of stress for both the child and the family in the long run. The best way to reduce this is to communicate properly with the family and to inform them that these situations are benign.

Limitations of the study were, as would be expected with any retrospective study. In a retrospective study, we were constrained by the information from the medical records.The results do not represent the actual incidence of NEPEs in the general population

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