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MULTI-PERSPECTIVE INVESTIGATION OF PAROXYSMAL NONEPILEPTIC EVENTS RETROSPECTIVELY

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Abstract

Non-epileptic paroxysmal events (NEPE) refer to physiological or exaggerated physiological responses caused by parasomnias, movement disorders, behavioral or psychiatric disorders and hemodynamic, respiratory and gastrointestinal dysfunctions. In the present study detailed data were provided on the demographic features of NEPE based on five-years of clinical experience and observation. The medical records of 200 patients diagnosed with NEPE were investigated retrospectively. The distribution of NEPE was evaluated based on age and sex. Our study demonstrated that 23.5% of patients with NEPE had previously been followed with a diagnosis of epilepsy. 34 (45.4%) patients aged 2 months to 4(11/12) years were diagnosed with breath holding spells. Psycogenic seizures (PS) was the most common diagnosis (32.5%) in general. The mean age in PS was 11.25 ± 3.42 years. 38 (58.5%) of them were female, and 27 (41.5%) were male. Some 28 (43.1%) PS were diagnosed from the amateur camera images taken by families; 20 (30.1%) were diagnosed from video-EEG monitoring; while the physician witnessed a seizure first-hand in 17 (26.2%) of the patients. The form and frequency of NEPE in children vary with age. PS was the most common diagnosis in accordance with the literature. In contradistinction to previous studies breath holding spell was found to be the most common NEPE in early childhood period for this study population.

Keywords: Non-epileptic paroxysmal events, Psycogenic seizure, Breath holding spell

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1. Introduction

Paroxysmal events are starting and end abruptly, characterized by altered consciousness, abnormal involuntary movements, and changes in muscle tone and breathing patterns. Paroxysmal events are not uncommon and can occur in all periods of childhood. While 70% of paroxysmal events in the first year of life are physiological and nonepileptic paroxysmal events (NEPE), the incidence of NEPE in older age groups range from 3.5 to 43% (Visser et al., 2010; Yalnızoglu et al., 2009; Montenegro et al., 2008).

The terms "convulsion" and "seizure" are used interchangeably terms for the identification of episodes involving involuntary motor activity. It should be kept in mind that it can be one of the physiological or exaggerated physiological responses related with NEPE, parasomnias, movement disorders, behavioral or

psychiatric disorders that the phenomenon described as seizure, rather than epilepsy. For an accurate diagnosis, appropriate treatment and good prognosis are to be ensured, it is necessary to distinguish NEPE from epilepsy. Non-epileptic events may exhibit stereotypical features and may mimic epileptic seizures (ES). Although there may be visible changes in motor and sensory functions, electroencephalography (EEG) may reveal no abnormal brain activity. If misdiagnosed with epilepsy, children may be exposed unnecessarily exposed to the side effects of antiepileptic drugs which may have negative effects on their social lives and academic achievements. Psychogenic seizures (PS) are a form of NEPE that occur as a result of a physiological process, and that cause no electrical brain activity, with the change in movement or feelings and have clinical characteristics that resemble those of ES. PS may be observed alone or concomitant to ES (Kutluay et al., 2010). Appropriate care for NEPE reduces inappropriate medical investigation and therapy, expedites rates of remission, and decreases health-care utilization in a pediatric setting (Sawchuk et al., 2015).

The frequency of NEPE in children is identified in literature from the results of video-EEG monitoring, although there is a lack of data about related to the demographic characteristics of NEPE by getting a diagnosis in daily clinical practice. In the present study, we provide detailed data on the demographic features of NEPE based on five years of clinical experience and observation.

2. Material and Method

The 497 patients diagnosed with NEPE that were admitted to the Ondokuz Mayıs University, Pediatric Neurology Clinic between August 2005 and February 2010 were reviewed retrospectively. The study included patients aged 2 months to 18 years who were diagnosed with NEPE according to international scientific criteria. A total of 200 patients who met the criteria were included in the study. Documentary information was recorded using a research form based on coding. The obtained data was then coded, and analyzed in SPSS (Statistical program in Social Sciences) 15.0. Descriptive data was expressed as mean ± standard deviation, median (smallest-largest), and number and percent.

3. Results

A total of 200 (40%) of the 497 patients diagnosed with NEPE were included in the study. The mean age was 7.7 ± 5.04 years. 106 (53.5%) of them were female and 94 (46.5%) were male. Some 45 of 200 (23%) patients had been followed up for epilepsy prior to these diagnosis of NEPE. The most common diagnosis was PS (32.5%) in general study population (Table 1). Breath holding spell was the second most common diagnosis (17.0 %).

Table 1. General diagnostic distribution of non-epileptic paroxysmal events

Diagnosis	n	%
Psychogenic seizures	65	32.5
Breath holding spell	34	17.0
Syncope	18	9.0
Stereotypes/Tics	15	7.5
Startle disease	14	7.0
Movement disorders	9	4.5
Sleep disorders	9	4.5
Masturbation	7	3.5
Gastroesophageal reflux	5	2.5
Benign paroxysmal vertigo	4	2.0
Shuddering attacks	4	2.0
Complicated migraine	4	2.0
Daydreaming	4	2.0
Cyclic vomiting	3	1.5
Extraocular eye movements	3	1.5
Alternan hemiplegia	1	0.5
Benign paroxysmal torticollis	1	0.5
Total	200	100,0

Some 34 (45.4%) patients aged 2 months to 4(11/12) years were diagnosed with breath holding spell (BHS) (Table 2). The second most common diagnosis (10.2%) in this group was startle disease. PS was the most common diagnosis both in the group of patients aged 5-11(11/12) years (43%) and 12-18 years (57%). In addition to them, syncope was the second most common diagnosis in both age groups (15 % and 14% respectively) (Table 3 and 4).

Table 2. Diagnostic distribution of patients aged 2 months-$4^{(11/12)}$ year

Diagnosis	n	%
Breath holding spell	34	45.4
Startle disease	8	10.6
Masturbation	6	8.0
Gastroesophageal reflux	5	6.6
Shuddering attacks	4	5.4
Psycogenic seizure	4	5.4
Benign paroxysmal vertigo	3	4.1
Sleep disorders	3	4.1
Stereotypes/Tics	2	2.6
Extraocular eye movements	2	2.6
Movement disorders	1	1.3
Benign paroxysmal torticollis	1	1.3
Cyclic vomiting	1	1.3
Daydreaming	1	1.3
Total	75	100

The mean age was 11.25 ± 3.42 years in PS. 38 (58.5%) of the respondents were female, and 27 (41.5%) were male. Some 28 (43.1%) of them were diagnosed from the amateur camera images taken by families; 20 (30.1%) were diagnosed from video-EEG monitoring; while the physician witnessed a seizure first-hand in 17 (26.2%) of the patients. A total of 30 (46.2%) of the cases were

followed up with for epilepsy prior to being diagnosed with PS. Seven (10.8%) had concomitant epilepsy; 29 (44.6%) had an accompanying psychiatric disorder; and five (7.7%) had a family member with epilepsy.

Table 3. Diagnostic distribution of patients aged 5-11(11/12) year

Diagnosis	n	%
Psycogenic seizure	32	43.0
Syncope	11	15.0
Stereotypes/Tics	9	12.2
Movement disorders	6	8.1
Sleep disorders	4	5.5
Daydreaming	3	4.0
Complicated migraine	2	2.2
Cyclic vomiting	2	2.7
Startle disease	2	2.7
Benign paroxysmal torticollis	1	1.4
Alternan hemiplegia	1	1.4
Masturbation	1	1.4
Total	74	100

Table 4. Diagnostic distribution of patients aged 12-18 year

Diagnosis	n	%
Psycogenic seizure	29	57
Syncope	7	14
Stereotypes/Tics	4	8
Startle disease	4	8
Movement disorders	2	4
Sleep disorders	2	4
Complicated migraine	2	4
Extraocular eye movements	1	1
Total	51	100

4. Discussion

This study showed 45/200 (23%) patients had been followed up for epilepsy prior to these diagnoses of NEPE. It was found that 39% of children who were diagnosed as having epilepsy were misdiagnosed, and approximately 47% of these diagnoses consisted of NEPEs in a previous study (Uldall et al., 2006).

It was suggested that although all patients were diagnosed by experienced child neurologists, nonepileptic events still may misdiagnosed as epilepsy. Diagnostic rate and understanding the semiologic characteristics of PS increased after the introduction of video EEG. In most studies, PS has already been shown to be the most common type of NEPE (Yalnızoglu et al., 2009; Kutluay et al., 2010; Yılmaz et al., 2013). PS was the most common diagnosis that 65 (32.5%) of the 200 patients were diagnosed with PS in the present study. Despite new developing technology, there may be still a delay 7.2 years on average between onset and the definite diagnosis of PS, as three- quarters of cases were diagnosed initially with epilepsy and were treated with anti-epileptic drugs (Wichaidit et al., 2015). However, it is a severe iatrogenic damage to start treatment for PS.

Some 10.8% of the PS patients had concomitant epilepsy. However this rate varies between 25-30% in other studies (Yalnızoglu et al., 2009; Kutluay et al., 2010). These patients with a double-diagnosis were following up only for epilepsy until a full video-EEG monitoring could be made. It was remarkable that these groups of patients showed heterogenity in their seizure semiology.

PS is categorized as dissociative disorder according to ICD-10, while in DSM-IV, it is referred to as somatoform or conversion disorders. The dominance of the female gender has been identified in several studies in adults (Benbadis and Chichkova, 2006; Asadi-Pooya and Sperling, 2015). On the other hand, the condition is related with age in children. Studies have shown that the majority of PS between 5-18 years are female (Bhatia and Sapra, 2005). PS was the most common diagnosis both in the group of patients aged 5-11(11/12) years (43%) and 12-18 years (57%). Although there was a female dominance in 12-18 years age group, no gender predominance was identified in the 5-11 (11/12) age group in the present study.

Many psychiatric disorders (somatization disorder, dissociative disorder, personality disorder, anxiety disorder, depression) have been defined in patients with PS. In a study of 50 PS patients, it was determined that 33 (46%) had concomitant psychiatric disorders (Tinuper et al., 2007). Some 44.6% of the PS patients in the present study had psychiatric disorders, similar to the findings in literature. There is a tendency to dissociative and somatization in both PS and other psychiatric disorders, although they have different intrapsychiatric mechanisms (Ristić et al., 2004). PS patients should be consulted and follow up should be synchronous with pediatric psychiatry if qualified treatment results are to be assured.

Home videos made on mobile phones are shown to be a cost-effective tool for the diagnosis of paroxysmal events. Mobile phone videos increased the mean accurate diagnoses by 11.5% for nonepileptic events (Huang et al., 2019). In most studies, the means of diagnosis of PS was via videoEEG monitoring. There have been few studies to date reporting on diagnoses based on the first-hand witnessing of a seizure by a clinician, or from watching the amateur videos of seizures taken by family members. In Syed et al. (2011)'s study of adult patients investigating semiological characteristics specific to PS, the data obtained from eyewitnesses was correlated with video images. In the present study, the diagnosis of 43.1% of the PS patients was made from amateur video recordings, accounting for a greater proportion of the diagnoses than video-EEG monitoring due to the fact that our video-EEG monitoring unit was not established until 2008. Although it may be considered an archaic and amateurish method, in the light of the obtained results, its usefulness is

incontrovertible when access to modern methods is impossible.

Breath-holding spells are nonepileptic paroxysmal phenomenon frequently seen in 6-48 month (Gürbüz et al., 2019). Although they can be confused with epileptic seizures and make the families frightened, they are benign conditions and generally resolve up to 4-5 years. In the present study, BHS was the most common diagnosis in patients aged 2 months-4 (11/12) years. Iron is known to have critical role for cerebral neurotransmission in addition to eritropoesis. Iron deficiency (ID) and ID anemia have been suggested as a predisposing factor in BHS (Mackay, 2005; Akçam, 2002; Gençgönül et al., 2002). Those aged between 6 months and 2 years are most likely common age period of having ID anemia. Likewise between these ages, ID anemia is too much common to be underestimated in our country (Şakru et al., 2000; Mocan et al., 1999). In a recent study, iron deficiency was detected 257 in 312 (82.4%) patients who have diagnosed with BHS (Gürbüz et al., 2019).

It is not challenging to make differential diagnosis in BHS if physician ask the accurate questions to families during the examination. Most of them describe the attacks occuring usually following an emotional stimulus or trauma. On the other hand, many families are anxious about the clinical presentation of spells. Consequently, this reason may explain the overmuch admission to the Pediatric Neurology Clinic as well as the high rate of BHS in the present study.

Vasovagal syncope; is the most common form of syncope in children. Vasogenic syncope (26.3%) was showed to be the second most frequently observed NEPE following PS in the adeolesan group (12 years<) in a study (Park et al., 2015). Similarly, syncope was the second most common diagnosis in the age group of 12-18 year (15%) as well as in the group of patients aged 5-11 (11/12) years (14%) in present study. Abnormal movements such as contractions of the extremities, occasionally accompanied by urinary incontinence, may ocur during syncope. In that case, it could be challenging to distinguish syncope from an epileptic seizure. Therefore, it is necessary to collaborate with pediatric cardiologist and performing a head-tilt test to go to the accurate diagnosis.

In the present study population, the frequencies of the sleep disorders were found to be low (4.5 %). However this rate is higher in the other studies (Kim et al., 2012; Kutluay et al., 2010). Although, the diagnoses of sleep disorders were done according to clinical experience of the physician, the patients reported previously have got diagnosis via video-EEG monitoring. It was suggested that this may explain the lower rates for sleep disorders in this study.

5. Conclusion

The incidence and frequency of NEPE in children varies according to age, but it should always be kept in mind as a possible diagnosis in epileptic patients who do not respond to treatment. The first step in making a differential diagnosis is to ensure there is sufficient knowledge and doubt about the subject. Subsequent diagnostic procedures should be followed to confirm the diagnosis. The optimum of diagnosis involves the physician witnessing a seizure first hand. As this is not always possible, the gold diagnostic method is video-EEG monitoring. Finding such a well-equipped monitoring room in most hospital may be difficult. If the event described by the family is not seen by the attending physician, the family may be asked to record a seizure on an amateur camera.

Conflict of interest

The authors declare that there is no conflict of interest.

Ethics

The study was approved by the ethics committee of Ondokuz Mayıs University (Document no: 2011/322).

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