Epilepsy & Behavior 96 (2019) 150–154

Contents lists available at ScienceDirect

Epilepsy & Behavior

journal homepage: www.elsevier.com/locate/yebeh

Presentation of psychogenic nonepileptic seizures in Hawaii's ethnoracially diverse population

Richard Ho [a], Jasen Ocol [a], Carol Lu [a], Shelby Dolim [a], Michael Yang [a], Enrique Carrazana [a], Kore Kai Liow [a,b,c,*]

[a] Epilepsy Research Unit, Hawaii Pacific Neuroscience, Honolulu, HI, USA
[b] Comprehensive Epilepsy Center and Epilepsy Monitoring Unit (EMU), Hawaii Pacific Neuroscience, Honolulu, HI, USA
[c] John A. Burns School of Medicine, University of Hawaii, Honolulu, HI, USA

ARTICLE INFO

Article history:
Received 22 March 2019
Revised 15 April 2019
Accepted 15 April 2019
Available online 28 May 2019

Keywords:
Video-EEG
Psychogenic
PNES
Native Hawaiian
Ethnicity
Semiology

ABSTRACT

Purpose: This exploratory study compared the semiology of psychogenic nonepileptic seizures (PNES) between a diverse group of patients in the state of Hawaii. This study may expand understanding of PNES across different ethnocultural and gender groups.
Methods: A retrospective chart review of patients admitted to our Epilepsy Monitoring Unit (EMU) over a 4-year period was performed to compare semiology in different ethnic groups and gender.
Results: A total of 139 patients were included in this study, 37% (n = 51) with PNES, 34% (n = 47) with epilepsy only, and 29% (n = 41) with other non-PNES, nonepilepsy diagnosis. The number of Asians with PNES were found to differ when compared with the patients with epilepsy and the patients with non-PNES, nonepilepsy diagnosis. A positive trend was found in the number of Native Hawaiians and Caucasians with PNES in comparison with patients with non-PNES, nonepilepsy diagnosis. In addition, three semiology of PNES in Native Hawaiians were found to differ in comparison with other ethnic groups with PNES: rhythmic motor, mixed semiology, and nonepileptic aura. There is a significant difference in all motor manifestation between males and females in Native Hawaiians. Between patients with PNES, patients with epilepsy, and patients with non-PNES, nonepilepsy diagnosis, significant correlation was found in psychiatric disorders including posttraumatic stress disorder (PTSD), anxiety, and any psychiatric disorder.
Conclusion: This cross-cultural study found significant differences in the expression of PNES across key ethnoracial groups for the Islands of Hawaii. These findings have implications to the diagnosis and treatment of PNES for Native Hawaiians and other Pacific Islanders in the United States.

1. Introduction

Patients with psychogenic nonepileptic seizures (PNES) tend to have common traumatic life experiences (e.g., physical or sexual abuse) that often lead to psychiatric comorbidities such as depression, anxiety, and posttraumatic stress disorder (PTSD) [1]. The expression of psychiatric disorders in relation to traumatic experiences varies among patients. It is often modulated by ethnoracial and cultural factors. These differences may suggest that there is a degree to which mental disorders and life trauma are somaticized [2]. Non-Caucasian, ethnoracial groups such as Hispanics and East Asians report a higher tendency for somatization as an expression of an underlying mental disorder. Gender and socioeconomic status may play a role as well [2,3]. Thence, it is reasonable to speculate that the prevalence and semiology of PNES may be different among various ethnoracial groups. As the population in the United States and other industrialized nations continues to become more diversified, data on the presentation of PNES among various ethnoracial groups are warranted.

Hawaii's diverse population provides a unique opportunity to explore PNES among different ethnicities and races (e.g., Asians and Native Hawaiians), which are often underrepresented in U.S.-Mainland-based studies. The purpose of this exploratory study was to investigate whether ethnoracial factors play a significant role in modulating the presentation of PNES among a diverse outpatient neurology practice in Hawaii.

2. Methods

A systematic retrospective review was conducted of patients referred to the Hawaii Pacific Neuroscience Epilepsy Monitoring Unit (EMU) for video-electroencephalogram (vEEG) for the exclusion of a seizure disorder as the diagnosis for their symptoms. The review included a four-year period (2014–2018) and was approved by an institutional review board. All charts and vEEG were reviewed by board-certified neurologists with epilepsy training (KL, EC). Patients without clear vEEG reports or incomplete testing were excluded from this

* Corresponding author at: St. Francis Medical Center, 2230 Liliha St #104, Honolulu, HI 96817, USA.
E-mail address: kliow@hawaii.edu (K.K. Liow).

https://doi.org/10.1016/j.yebeh.2019.04.024
1525-5050/

retrospective chart review. Patients' demographics and psychiatric history were captured. Also, patients were characterized into racial and ethnocultural subgroups based on self-identification: Caucasian, Asian, Native Hawaiian, other, or unreported. Upon analysis of vEEG, patients' with PNES manifestations of their sudden episodes were categorized as rhythmic motor, hypermotor, complex motor, dialeptic, nonepileptic aura, and mixed presentations. This classification scheme along with details on the semiologic features of each category was developed by Seneviratne and colleagues based upon clinical observation of vEEG-confirmed PNES episodes [4].

Descriptive and bivariate statistical analysis was performed multiple times to compare different factors. First, chi-square test was conducted to compare demographics between the patients with PNES, patients with epilepsy, and patients with other non-PNES, nonepilepsy diagnosis. Second, chi-square analysis was utilized to compare ethnicity, psychiatric disorders (depression, anxiety, PTSD, and any psychiatric disorder), and marital status. Third, chi-square analysis was conducted to determine association between PNES semiology/ethnicity and PNES semiology/gender. Further analysis was performed in PNES semiology/gender with ethnic groups, specifically Native Hawaiian and Caucasian. Finally, chi-square analysis was used to determine the association between psychological disorders and gender in the patients with PNES and ethnic groups with PNES. All statistical analysis was performed using IBM SPSS 25.0 statistical software. As this was an exploratory study, p-values are quoted uncorrected for multiple comparisons. A p-value of 5% or less was used to define significance, and a p-value of less than 10% was identified as a trend.

3. Results

A total of 139 patients were included in the study. Based upon clinical observation and vEEG diagnostic examinations of the episodes along with medical records, 51 patients were identified with PNES. Within the patients with PNES, there were 19 patients with coexisting epilepsy. Of the remaining 88 patients, vEEG conclusively diagnosed epilepsy in 47 patients. Forty-one patients were determined to have other non-PNES, nonepilepsy diagnosis such as complex migraine, transient ischemic attack, and cardiovascular syncope. This group of patients was referred to the EMU for vEEG to exclude epileptic seizures as the etiology for their paroxysms.

Table 1 summarizes the demographic characteristics of patients with PNES in comparison with patients with epilepsy and patients without epilepsy nor PNES. Chi-square analysis showed that there was a significant difference in gender in association with PNES in comparison with patients with epilepsy. There was also a positive trend in the number of Native Hawaiians and Caucasians with PNES in comparison with patients without PNES or epilepsy. The number of Asians with PNES also differed in comparison with patients with epilepsy and patients without PNES or epilepsy. Moreover, some psychiatric disorders were found to have marginal to significant correlation between patients with PNES, patients with epilepsy, and patients without epilepsy or PNES, specifically PTSD, anxiety, and any psychiatric disorders. However, no significant difference was found in rates of depression in patients with PNES. There was also no significant difference found in marital status.

In comparison with the other ethnic groups with PNES, Native Hawaiians have a significant difference in presentation with respect to nonepileptic aura and a trend for rhythmic motor and mixed semiology presentations. Other semiology (summarized in Table 2) showed no significant differences between ethnic groups.

Subsequent analysis was performed to test our hypothesis if diversification of PNES semiology can be found across gender and the two predominant ethnic groups with PNES. As shown in Table 3a, the semiology of PNES between males and females was compared. There was a marginal significance found in mixed semiology between males and females ($X2 = 3.709$; $p = 0.054$). The semiology of Caucasians and Native Hawaiians was compared in Table 3b. There was also a marginal significant difference in all motor manifestation between males and females in Native Hawaiians ($X2 = 4.098$, $p = 0.043$). However, no significant differences were found in other semiology between gender in Native Hawaiians and Caucasians.

Further analysis was performed to test if psychological disorders differ between gender and ethnic groups with PNES. In Table 4a, the number of patients with PNES with depression was significant when comparing between males and females. Females were more likely to have depression. No significance was seen in other psychological

Table 1
Comparison of demographic characteristics of patients with PNES, patients with epilepsy, and patients without epilepsy.

	Total sample ($n = 139$)	Patients with PNES ($n = 51$)	Patients with epilepsy only (n = 47)	Patients with other diagnosis (n = 41)
Gender		****(p = 0.0016)**		
Male *n* (%)	65 (46.8)	18 (35.3)	28 (59.6)	19 (46.3)
Female *n* (%)	74 (53.2)	33 (64.7)	19 (40.4)	22 (53.7)
Age mean (SD: standard deviation)	49 (18.6)	44 (16.7)	49 (18.1)	56 (19.7)
Ethnicity				
Native Hawaiian *n* (%)	34 (24.5)	17 (33.3) *(p = 0.078)*	10 (21.3)	7 (17.1)
Caucasian *n* (%)	35 (25.2)	16 (31.4) *(p = 0.061)*	13 (27.7)	6 (14.6)
Asian *n* (%)	24 (17.3)	4 (7.8) **(p = 0.099)** **(p = 0.014)*	9 (19.1)	11 (26.8)
Other ethnicity *n* (%)	18 (12.9)	9 (17.6)	3 (6.4)	6 (14.6)
Unreported *n* (%)	28 (20.1)	5 (9.8)	12 (25.5)	11 (26.8)
Marital status				
Married *n* (%)	49 (35.3)	16 (31.4)	19 (40.4)	14 (34.1)
Single/divorced/separated *n* (%)	90 (64.7)	35 (68.6)	28 (59.6)	27 (65.9)
Psychiatric disorder				
Depression	57 (41.0)	24 (47.1)	19 (40.4)	14 (34.1)
PTSD	11 (7.9)	8 (15.7) **(p = 0.062)** **(p = 0.034)*	2 (4.3)	1 (2.4)
Anxiety	37 (26.6)	24 (47.1) ****(p = 0.001)** ***(p = 0.002)*	6 (12.8)	7 (17.1)
Any psychiatric disorder	70 (50.4)	32 (62.7) ***(p = 0.045)** *(p = 0.071)*	20 (42.6)	18 (43.9)

Note: Bold represents PNES vs. epilepsy; italics represents PNES vs. other diagnosis.
* $p \le 0.05$.
** $p \le 0.01$.

Table 2
Semiology of patients with PNES in different ethnic groups.

	Total (n = 51)	Native Hawaiian (n = 17)	Caucasian (n = 16)	Asian (n = 4)	Other/unreported ethnicity (n = 14)
Rhythmic motor n (%)	6 (11.8)	4 (23.5) (p = 0.065)	2 (12.5)	0 (0.0)	0 (0.0)
Hypermotor n (%)	5 (9.8)	1 (5.9)	2 (12.5)	1 (25.0)	1 (7.1)
Complex motor n (%)	7 (13.7)	3 (17.6)	2 (12.5)	0 (0.0)	2 (14.3)
All motor manifestation n (%)	18 (35.3)	8 (47.1)	6 (37.5)	1 (25.0)	3 (21.4)
Dialeptic n (%)	11 (21.6)	3 (17.6)	3 (18.8)	2 (50.0)	3 (21.4)
Nonepileptic aura n (%)	16 (31.4)	2 (11.8) *(p = 0.033)	6 (37.5)	1 (25.0)	7 (50.0)
Mixed n (%)	6 (11.8)	4 (23.5) (p = 0.065)	1 (6.3)	0 (0.0)	1 (7.1)

Notes: Comparisons are made between each ethnic group using the total group of patients with PNES as the reference group. X2 analyses were conducted.
* p ≤ 0.05.

disorders between genders, such as PTSD and anxiety. In Table 4b, a trend was found for depression between Native Hawaiians and the group with PNES (X2 = 3.188, p = 0.074). However, no significance was seen in other psychological disorders across Native Hawaiians with PNES and their Caucasian counterparts.

4. Discussion

There is a limited number of studies which explore differences in semiology among groups of patients on the basis of ethnoracial and cultural differences. In a recent study comparing patients with PNES from Canada, Iran, and Saudi Arabia, a higher rate of preictal auras characterized as autonomic features of anxiety, such as hyperventilation and chest pain, was reported by Canadian patients. However, the authors felt that this was due to differences in the clinical assessment instead of differences in PNES semiology among patients [5]. In a separate study, American patients more often reported subjective seizures than their Brazilian counterparts [6]. The clinical and historical aspects of the cases were not significantly different among Brazilians and Americans; however, the groups were not further classified by their ethnoracial background. Surprisingly, only minor differences in presentation were reported in these two studies [5,6] despite the strikingly different cultural backgrounds among the groups. In a large cohort of patients from the Southeastern United States, a nearly twofold larger period prevalence of temporal lobe epilepsy (TLE) among African-Americans over non-Hispanic Caucasians patients was observed. Yet, no significant differences were noted in the prevalence of PNES among the two racial groups [7].

The low number (7.8%) of Asian-Americans presenting with PNES at our EMU contrasts not only with the comparator cohorts (19.1%, 26.8%), but also with the U.S. Census Bureau statistics for the State of Hawai'i in 2017 in which Asians represented 37.7% of the State's population [8]. Others have reported that Asian-Americans, as a group, endorse less affective symptomatology than other American ethnoracial groups, even when demographic variables such as gender and socioeconomic status are accounted for [9–12]. Higher education, stronger social support, and lower rates of substance abuse disorders have been speculated to contribute to lower psychological traumatic experiences exposure in Asian-Americans [11]. The number of Native Hawaiian with PNES was strikingly higher than expected from the U.S. Census Bureau statistics for the State of Hawaii (33.3% vs. 10.2%) [8]. While there was a trend for higher representation of the Native Hawaiian ethnicity among the group with PNES versus their epilepsy counterparts, similar percentages of Native Hawaiians across all three groups suggest that these observations could be related to our epilepsy center patient referral patterns.

A number of PNES semiology classification systems has been proposed [4,6,13]. A recently published classification system distills the seizure types into 4 groups by focusing on the most prominent seizure characteristics [6,13]. We opted to apply the semiology classification proposed by Seneviratne and colleagues as it provides additional segmentation of motor manifestations and mixed patterns, taking into consideration that the aim of our study was to identify potential differences in the expression of PNES among the major ethnoracial groups in Hawaii. This system was derived from a systematic vEEG monitoring of a significant number of patients with PNES and their episodes. Episodes were classified into six groups: rhythmic motor, hypermotor, complex motor, dialeptic (prolonged unresponsiveness), nonepileptic auras, and mixed [4]. In our cohort, Caucasians with PNES presented more often with nonepileptic auras, while Native Hawaiians tended to present with more expressive rhythmic motor or mixed manifestations for their episodes. This difference in the PNES semiology of the Native Hawaiian patients may be shaped by differences in the reaction to psychological trauma. Native Hawaiians have been observed to report more physical symptoms in relation to trauma exposure than other ethnoracial groups in Hawaii [14,15].

In concordance with prior reports, there was a female preponderance among our subjects in comparison with the epilepsy group (64.7% vs. 40.4%, p = 0.0016). It has been suggested that an elevated risk of sexual abuse contributes to PNES manifesting more often in women [16,17]. Additionally, previous literature has reported that women endorse significantly higher rates of sexual trauma,

Table 3a
Semiology of PNES between males and females.

	Male (n = 18)	Female (n = 33)
Rhythmic motor n (%)	2 (11.1)	4 (12.1)
Hypermotor n (%)	3 (16.7)	2 (6.1)
Complex motor n (%)	3 (16.7)	4 (12.1)
All motor manifestation n (%)	8 (44.4)	10 (30.3)
Dialeptic n (%)	4 (22.2)	7 (21.2)
Nonepileptic aura n (%)	6 (33.3)	10 (30.3)
Mixed n (%)	0 (0.0)	6 (18.2)

Table 3b
Semiology of PNES between Native Hawaiian males and females, Caucasian males and females.

	Caucasian (n = 16)		Native Hawaiian (n = 17)	
	Male (n = 7)	Female (n = 9)	Male (n = 3)	Female (14)
Rhythmic motor n (%)	1 (14.3)	1 (11.1)	1 (33.3)	3 (21.4)
Hypermotor n (%)	1 (14.3)	1 (11.1)	1 (33.3)	0 (0.0)
Complex motor n (%)	1 (14.3)	1 (11.1)	1 (33.3)	2 (14.3)
All motor manifestation n (%)	3 (42.9)	3 (33.3)	3 (100.0)*	5 (35.7)*
Dialeptic n (%)	1 (14.3)	2 (22.2)	0 (0.0)	3 (21.4)
Nonepileptic aura n (%)	3 (42.9)	3 (33.3)	0 (0.0)	2 (14.3)
Mixed n (%)	0 (0.0)	1 (11.1)	0 (0.0)	4 (28.6)

* p ≤ 0.05.

Table 4a
Psychological disorders between male and female patients with PNES.

	Male patients with PNES (n = 18)	Female patients with PNES (n = 33)	X2	p value
Psychological disorder				
Depression	5	19	4.151	0.042*
PTSD	2	6	0.440	0.507
Anxiety	9	15	0.097	0.756
Any psychiatric disorder	10	22	0.615	0.433

* $p \leq 0.05$.

dissociations, and sexual disturbances while men report a higher use of avoidance and depressive symptoms [18]. Similarly, higher rates of PTSD secondary to sexual trauma have been found in women with coexistent PNES and refractory TLE [19].

The significant correlation and trends between psychiatric disorders (i.e., depression, PTSD, and anxiety) and PNES noted in our study (Table 1) were expected, as traumatic life experiences and psychiatric comorbidities are common among patients with PNES [1]. Moreover, compared with men, women with PNES had more psychiatric diagnosis, mainly depression, confirming previous studies that have examined PNES and gender in terms of these variables [19,20] (Table 4a). No significant differences were noted among genders when examined across the two most represented ethnoracial groups, Native Hawaiians and Caucasians (Table 4b). Larger numbers will be needed in subsequent studies to confirm this observation. The data may suggest that factors tied to the ethnic and racial make-up of the patients might be modulating the expression of PNES.

The limited numbers of studies that have specifically looked at gender differences in semiology have suggested that there are no major differences in adults [16,17,20,21] or in adolescents [22] with PNES. Episodes in males tend to be brief in duration [20], manifest at younger ages [20], and present with motor manifestations [21]; however, in our study, women with PNES tended to present with a mixed semiology more so than their male counterparts. While the numbers are small, the majority of these patients reported Native Hawaiian ethnicity. This study does not corroborate prior suggestion that PTSD related to sexual trauma has a modest association with more severe and complex PNES episodes [23].

Our study has limitations worthy of discussion. While the number of patients included in this exploratory study is in line with other recently published series of patients with PNES [5,6,19,20], the addition of further patients is required to confirm our observations with more certainty. Patients were derived from a single EMU; thus, the study may be sensitive to patient referral patterns and not be fully representational of all PNES and patients with epilepsy in Hawaii. In addition, as the data were retrospectively collected, we are unable to further assess the patients' psychiatric history in more detail than provided in their comprehensive medical records. Lastly, a self-identification by patients was implemented to define ethnoracial groups. Ethnicities such as "Hawaiian" may include a diverse mixed ethnoracial background given that it refers to a person with Hawaiian ancestry regardless of blood quantum. The dilemma of self-classification into a specific ethnoracial group for individuals of mixed family backgrounds might explain the 20% of all patients who did not report race or ethnic group. Similarly, the U.S. Census Bureau reports that 23.8% of individuals in Hawaii self-report to be of two or more races [8].

Table 4b
Psychological disorder in different ethnic groups with PNES.

	Total (n = 51)	Caucasian (n = 16)	Native Hawaiian (n = 17)	Asian (n = 4)
Psychological disorder				
Depression	24	8	11	0
PTSD	8	3	3	0
Anxiety	24	8	7	1
Any psychiatric disorder	32	10	13	1

To conclude, this exploratory study, first for Hawaii, found possible differences in the expression of PNES across key ethnoracial groups for the Islands. Corroboration of such differences in subsequent studies does have implications on the diagnosis and treatment of PNES for the 1.3 million Native Hawaiians and other Pacific Islanders living across the United States [24]. Moreover, future studies on PNES might want to consider looking further into ethnoracial factors and their relation to presentations and treatment outcomes.

Acknowledgments

This was an unfunded study. The authors thank all the patients and staff at Hawaii Pacific Neuroscience for their help and support. We also thank Catherine Mitchell for her editorial input.

Conflict of interest

The authors have no conflicts of interest to declare with regard to this publication.

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