Epilepsy & Behavior Case Reports 10 (2018) 129–132

Contents lists available at ScienceDirect

Epilepsy & Behavior Case Reports

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

Case Report

Recurrent focal seizures as a feature of status epilepticus presenting as a peri-ictal water drinking

Shuo Huang [a], Haifa Al-Abri [b], Alok Sachdeva [c], Ayham M. Alkhachroum [d], Stephanie Shatzman [b], Hans Lüders [b,*]

[a] Department of Neurology, The First Affiliated Hospital of Harbin Medical University, Harbin, China
[b] University Hospitals - Cleveland Medical Center, Department of Neurology, Case Western Reserve University School of Medicine, Cleveland, OH, USA
[c] Michael Aldrich Sleep Disorders Center, Department of Neurology, University of Michigan Health System, Ann Arbor, MI, USA
[d] Neurocritical Care, New York-Presbyterian, Columbia University Medical Center, New York, NY, USA

ARTICLE INFO

Article history:
Received 18 August 2018
Received in revised form 4 September 2018
Accepted 5 September 2018
Available online 9 October 2018

Keywords:
Peri-ictal water drinking
Status epilepticus
Seizure

ABSTRACT

We report a case of focal status epilepticus (SE) associated with peri-ictal water drinking (PIWD) behavior in a nine-year-old left-handed boy with epilepsy. We reviewed prior cases of epileptic peri-ictal water drinking. Only one adult patient with status epilepticus and PIWD has been reported previously. This is the first reported case of PIWD SE in a pediatric patient with frontal lobe epilepsy. We found PIWD to have no lateralizing value.

1. Introduction

Peri-ictal autonomic signs and symptoms have been described in both adult and childhood epilepsy [1,2]. Many have been described, including cardiovascular, respiratory, gastrointestinal, and cutaneous manifestations [1–3]. Peri-ictal water drinking (PIWD) is a rare autonomic symptom, and along with others is commonly observed in patients with temporal lobe epilepsy (TLE) [2]. PIWD has been defined as the urge to drink water during a seizure or up to 2 min post-ictally [4].

Lennox and Cobb were the first to publish a report of PIWD in 1933 and later Rémillard described a patient with epilepsy who always carried a hip-flask of water and stated that water aborts his seizures [5].

2. Material and methods

We reviewed 88 electrographic seizures during 49 h and 39 min of video electroencephalogram (EEG) recording of a nine-year-old boy to determine whether electrographic seizures correlated with PIWD. We also monitored the R–R interval of the EKG 120 s before seizure onset and assessed whether there was a correlation between the EEG seizure and changes in heart rate. We defined ictal tachycardia as a 20% increase in HR from baseline. The baseline HR was calculated as the mean HR during the 120 to 300 s preceding electrographic seizure onset.

* Corresponding author at: University Hospitals - Cleveland Medical Center, Department of Neurology, 11100 Euclid Avenue, Cleveland, OH 44106-5040, USA.
E-mail address: Hans.Luders@UHhospitals.org (H. Lüders).

3. Results

3.1. Case report

A nine-year-old left-handed developmentally normal boy presented to the Pediatric Epilepsy Monitoring Unit (PMU) after suffering at least 8 h of confusion, word-finding difficulty, and decreased activity level without return to baseline.

At 8 years old, the patient complained of an abnormal sensation in his stomach and a feeling of thirst, which was followed by a ten-minute period of confusion. A routine EEG completed at that time was normal. Three months prior to presentation, he had an episode of thirst, confusion and word-finding difficulty, which was followed by unusual posturing of the fingers of his right hand and rightward head turning; these symptoms lasted 5 to 10 min, then progressed to a generalized tonic–clonic seizure. Patient was admitted to the Pediatric Intensive Care Unit (PICU) and was later discharged on levetiracetam (LEV). One month later, the patient was readmitted to a PICU with status epilepticus, at which point his LEV dose was increased. Video-EEG showed spikes in the left fronto-temporal region as well as continuous slowing in the left hemisphere. Magnetic resonance imaging (MRI) and Positron emission tomography (PET) are demonstrated in Fig. 1.

The patient was admitted to the PMU and monitored on video-EEG for 49 h and 39 min, during which time 88 electrographic seizures were recorded. EEG seizure duration was between 10 and 274 s with a mean of 78.2 s (SD 48.2 s). The time to clinical seizure onset after EEG

https://doi.org/10.1016/j.ebcr.2018.09.004

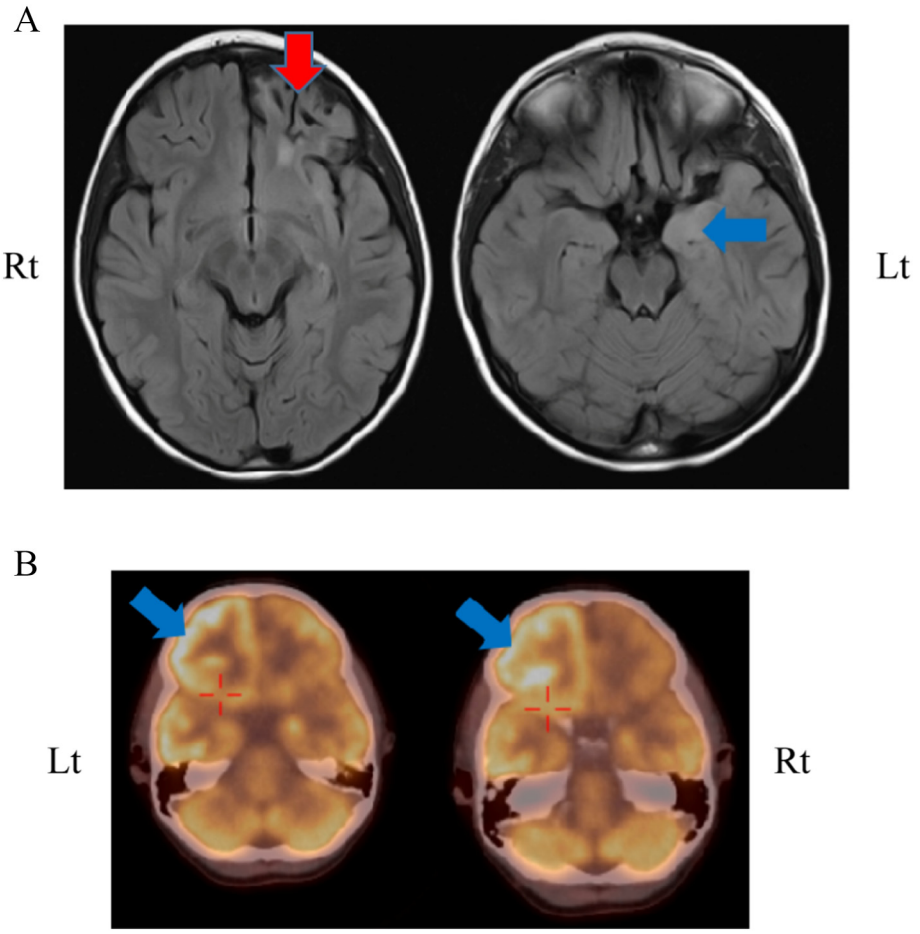

Fig. 1. (A) Brain MRI. Encephalomalacia and gliotic changes involving the left frontal lobe (Red arrow). Mild increased FLAIR signal involving the left hippocampus without evidence of tissue loss (Blue arrow). (B) PETCT. Increased metabolic activity within the inferior left frontal lobe (Blue arrows) corresponding to the area of encephalomalacia seen on MRI.

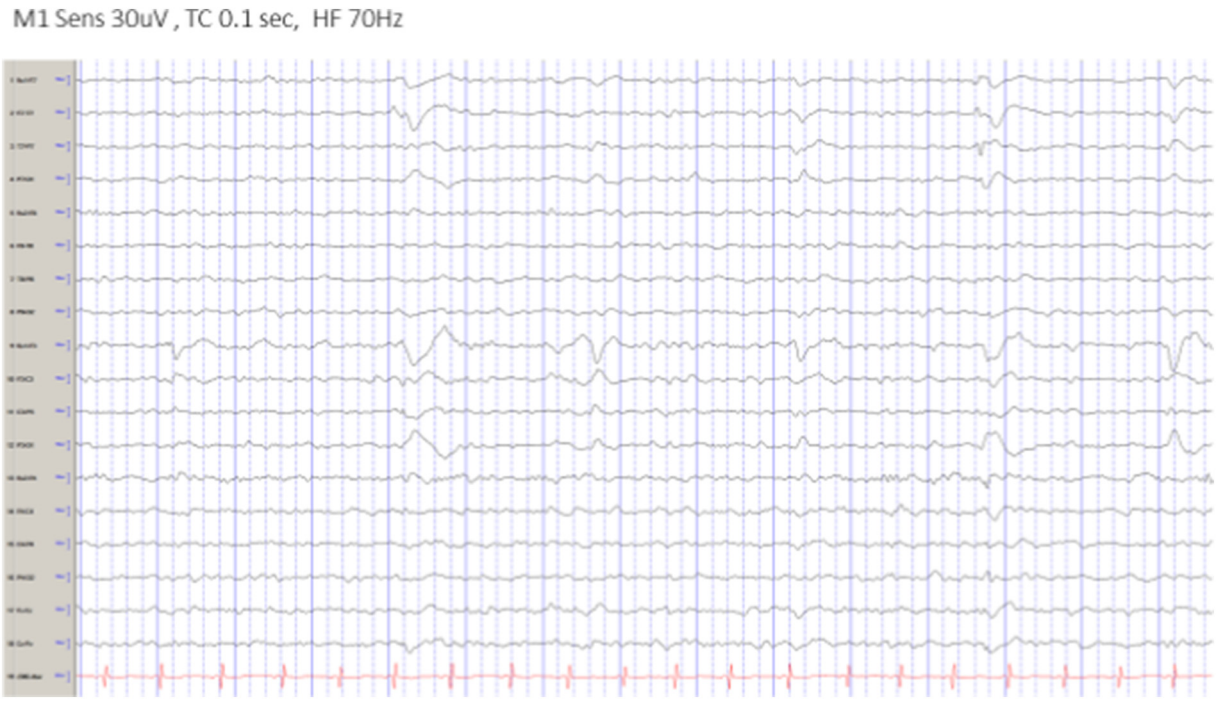

Fig. 2. Interictal EEG: interictal sharp waves in the left frontal and left temporal regions.

onset was variable, and ranged from 5 to 118 s. EEG at time of admission showed left frontal focal status epilepticus. Interictal epileptiform activity consisted of lateralized periodic discharges (LPDs) maximal either at F3 or at C3 (Fig. 2). Seizure's examples are shown in Fig. 3. The EEG seizures lasted between 10 and 274 s. Focal status epilepticus persisted for approximately 24 h. Seizures' localization is demonstrated in Table 1. Clinical features of seizures are shown in Table 2.

22 recorded seizures (25%) were associated with PIWD: the patient had water during 21 seizures and asked for water once. Out of the 22 seizures, 18 episodes of PIWD occurred during an electrographic seizure

Table 1
Distribution of seizure onset.

Maximum localization	Sleep stage	Awake stage	Amount (%)
F3	63	6	69 (78.4%)
T7	4	7	11 (12.5%)
C3	3	0	3 (3.4%)
P3	2	0	2 (2.3%)
Fp2	2	0	2 (2.3%)
P7	1	0	1(1.1%)
Amount (%)	75 (85%)	13 (15%)	88 (100%)

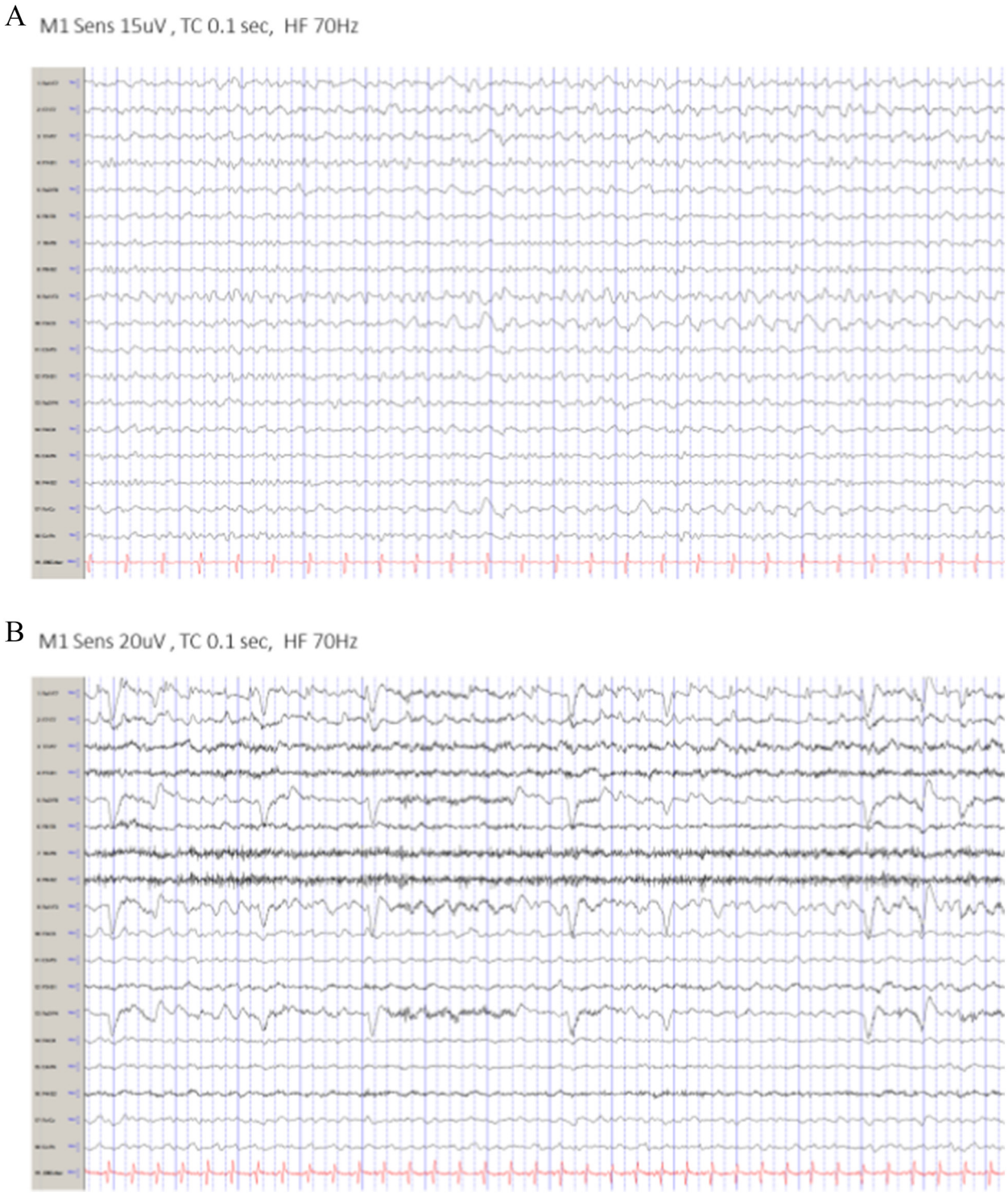

Fig. 3. (A) Ictal EEG onset with rhythmic discharges in the left hemisphere, maximum at F3. (B) At the beginning of ictal water drinking, there is rhythmic activity over the left hemisphere.

Table 2
Clinical features of EEG seizures.

Stage	EKG	Semiology
Sleep: 75 (85.2%)	EKG change: 59 (67.1%)	Drinking water: 10 (11.4%) Arousal: 22 (25%) No clinical signs: 27 (30.7%)
	No EKG change: 16 (18.2%)	Waking up: 1 (1.1%) Arousal: 1 (1.1%) No clinical signs: 14 (15.9%)
Awake: 13 (14.8%)	EKG change: 12 (13.7%)	Drinking water: 11 (12.5%) No clinical signs: 1 (1.1%)
	No EKG change: 1 (1.1%)	Drinking water: 1 (1.1%)

while four episodes occurred post-ictally, ranging between 8 and 102 s after the end of a seizure. In 10 out of 22 seizures with PIWD seizures, drinking or asking for water was the only clinical sign. During 12 episodes of PIWD, the patient experienced concurrent epigastric discomfort, somatosensory aura (nose itching), fear, and confusion.

4. Discussion

We report the case of a nine-year-old left-handed boy with left frontal focal status epilepticus associated with water-drinking behavior. To our knowledge, this is the first report of PIWD in a child with status epilepticus.

To date, there are 64 cases of seizures associated with PIWD [4,5]. The prevalence is low of PIWD with average age of 19 years old (SD 20), predominantly in women (66%). PIWD was observed predominantly during the ictal phase, but postictal and ictal/postictal have also been reported.

There are five possible mechanisms for PIWD: (1) thirst aura; (2) drinking to arrest a seizure; (3) drinking as an automatism; (4) dysgeusia (i.e. gustatory aura) and (5) disturbance of tissue osmolality [4–6]. In our case, three seizures were associated with a feeling of thirst and there was no evidence that drinking was triggered by dry mouth or dysgeusia. Serum electrolytes, serum and urine osmolality, and fasting blood glucose were all normal. Our patient never drank water before a seizure and there is no evidence that water-drinking arrested seizure activity. When water was not accessible during a seizure, some patients experienced extreme discomfort leading to accidental injury [7].

Mesial temporal seizures can propagate through the fornix, preoptic nucleus and anterior hypothalamus, which may result in water drinking even in the absence of high osmolality or hyperthermia. Mogenson and Stevenson showed that electrical stimulation of the lateral hypothalamus triggers water drinking [8,9]. This evidence suggests that ictal water drinking in patients with temporal lobe epilepsy may be due to abnormal excitation of the hippocampus spreading to the hypothalamus through the fornix. PIWD is most likely not a reliable lateralizing symptom [2,5,10]. This also explains the autonomic disturbance that patients may have, commonly sinus tachycardia [11]. Most of our patient's seizures provoked sinus tachycardia, which usually occurred after EEG seizure onset. This suggests that, the symptomatogenic zone of ictal tachycardia and PIWD involves the hypothalamus. Most prior cases of PIWD have been reported in patients with temporal lobe epilepsy [2,4,5,7,10,12]. Rémillard et al. used depth electrodes to show that seizures associated with PIWD began in the left amygdala and hippocampus, further supporting the theory of temporal lobe localization [5]. In our case, however, all the evidences point to a left frontal epileptogenic zone but the PIWD is due to spread of the epileptiform activity into the left temporal lobe.

Some pathologies associated with PIWD include mesial temporal sclerosis [13], post-infectious [5], autoimmune paraneoplastic encephalitis [6], vascular lesions such as cavernoma [10] and tumors [6,10]. Some cases underwent a surgical treatment [6,7,14].

5. Conclusion

We describe the clinical and electrographic features of a pediatric patient with PIWD due to status epilepticus arising from the left frontal region. To our knowledge, this is the first report of PIWD in a child with status epilepticus. PIWD most likely is of temporal lobe origin without lateralizing value.

Financial disclosure

Authors have no financial disclosure to report.

Conflict of interest

Authors report no conflict of interest. All authors agree to the journal's terms. The paper is not being submitted for publication elsewhere. The work is original and not published elsewhere. The manuscript meets the journal guidelines in ethics of publishing.

References

[1] Fogarasi A, Janszky J, Tuxhorn I. Autonomic symptoms during childhood partial epileptic seizures. Epilepsia 2006;47:584–8.
[2] Janszky J, Fogarasi A, Toth V, Magalova V, Gyimesi C, Kovacs N, et al. Peri-ictal vegetative symptoms in temporal lobe epilepsy. Epilepsy Behav 2007;11:125–9.
[3] Baumgartner C, Lurger S, Leutmezer F. Autonomic symptoms during epileptic seizures. Epileptic Disord 2001;3:103–16.
[4] Trinka E, Walser G, Unterberger I, Luef G, Benke T, Bartha L, et al. Peri-ictal water drinking lateralizes seizure onset to the nondominant temporal lobe. Neurology 2003;60:873–6.
[5] Rémillard GM, Andermann F, Gloor P, Olivier A, Martin JB. Water-drinking as ictal behavior in complex partial seizures. Neurology 1981;31:117–24.
[6] Pietrafusa N, Trivisano M, de Palma L, Serino D, Moavero R, Benvenga A, et al. Peri-ictal water drinking: a rare automatic behaviour in temporal lobe epilepsy. Epileptic Disord 2015;17:384–96.
[7] Cascino GD, Sutula TP. Thirst and compulsive water drinking in medial basal limbic epilepsy: an electroclinical and neuropathological correlation. J Neurol Neurosurg Psychiatry 1989;52:680–1.
[8] Mogenson GJ, Gentil CG, Stevenson JA. Feeding and drinking elicited by low and high frequencies of hypothalamic stimulation. Brain Res 1971;33:127–37.
[9] Mogenson GJ, Stevenson JA. Drinking induced by electrical stimulation of the lateral hypothalamus. Exp Neurol 1967;17:119–27.
[10] Szucs A, Fogarasi A, Rásonyi G, Kelemen A, Narula L, Tóth V, et al. Peri-ictal water drinking in temporal lobe epilepsy: is it a reliable lateralizing sign? Epilepsy Behav 2007;11:578–81.
[11] Sevcencu C, Struijk JJ. Autonomic alterations and cardiac changes in epilepsy. Epilepsia 2010;51:725–37.
[12] Crapanzano KA, Casanova MF, Toro VE, Gallagher B. Drinking behavior as a result of a right hippocampal ictal focus. Biol Psychiatry 1993;34:889–92.
[13] Musilová K, Kuba R, Brázdil M, Tyrlíková I, Rektor I. Occurrence and lateralizing value of "rare" peri-ictal vegetative symptoms in temporal lobe epilepsy. Epilepsy Behav 2010;19:372–5.
[14] Errguig L, Lahjouji F, Belaidi H, Jiddane M, Elkhamlichi A, Dakka T, et al. Peri-ictal water drinking and other ictal vegetative symptoms: localizing and lateralizing the epileptogenic zone in temporal lobe epilepsy? Two case reports and review of the literature. Rev Neurol (Paris) 2013;169:903–10.