

# Peri-ictal water drinking: a rare automatic behaviour in temporal lobe epilepsy

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**ABSTRACT** – Peri-ictal water drinking (PIWD) has been reported as the action of drinking during or within two minutes of an electroclinical seizure. It is considered a peri-ictal vegetative symptom, evident both during childhood and adulthood epilepsy. The aim of this paper was to describe the clinical and electroencephalographic features of two new adult subjects suffering from symptomatic temporal lobe epilepsy with episodes of PIWD recorded by VIDEO-EEG and to review literature data in order to better define this peculiar event during seizures, a rare and probably underestimated semiological sign. To date, 51 cases with focal epilepsy and seizures associated with PIWD have been reported. All patients presented with temporal lobe epilepsy. All cases but one had symptomatic epilepsy. Most of the patients had an involvement of the right hemisphere. Water drinking was reported as an ictal sign in the majority of patients, and less frequently was reported as postictal. We believe that PIWD might be considered a rare automatic behaviour, like other automatisms. Automatisms are more frequently described in patients with temporal lobe epilepsy. PIWD was reported also to have lateralizing significance in the non-dominant temporal lobe, however, because of its rarity, this finding remains unclear.

**Key words:** peri-ictal drinking, temporal lobe epilepsy, epilepsy/seizure and drinking/thirst, temporal lobe epilepsy

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Drinking behaviour is controlled by information from peripheral receptors processed in the preoptic area, the organum vasculosum of the lamina terminalis, the median preoptic nucleus, and the subfornical organ of the hypothalamus (Denton *et al.*,

1996). Moreover, the hippocampus and the amygdala are connected to the hypothalamus by a variety of multisynaptic reciprocal pathways that have been implicated in water balance and drinking behaviour (Gloor, 1997).

While the osmoreceptive role of these brain regions has been established, the anatomical network involved in thirst is not clear. Neuroimaging studies, however, showed that also cortical areas play a key role in the thirst mechanism. Activation during functional MRI has been reported for the anterior cingulate, parahippocampal gyrus, inferior and middle frontal gyri, insula, striatum and cerebellum. Furthermore, PET revealed significant activations observed in the anterior and posterior cingulate, parahippocampal and orbital frontal gyri, insula, claustrum, thalamus, and cerebellum (Denton *et al.*, 1999).

Drinking has been reported as an ictal sign in epileptic seizures (Rémillard *et al.*, 1981; Trinka *et al.*, 2003; Janszky *et al.*, 2007; Szucs *et al.*, 2007; Musilova *et al.*, 2010). Epileptiform activity generated in the temporal lobe may propagate into the hypothalamus and other structures involved in thirst and water regulation, producing ictal manifestations of abnormal water-seeking behaviour.

Peri-ictal water drinking (PIWD) was described as the action of drinking during or within two minutes of an electroclinical seizure (Trinka *et al.*, 2003). It is considered a peri-ictal vegetative symptom, evident both during childhood and adulthood epilepsy.

PIWD was firstly described in the early 1900s by Lennox and Cobb (1933), but it has been largely underestimated because drinking water *per se* is a very common event, and rarely patients or witnesses recognize it as part of an epileptic seizure. Different reports have been published describing PIWD in patients affected by temporal lobe epilepsy, mainly of symptomatic aetiology. In some of them, a possible lateralizing value has been hypothesized (Trinka *et al.*, 2003; Musilova *et al.*, 2010).

The recognition and interpretation of this ictal sign may be important to better define the epileptic syndrome, which was originally described in epilepsy surgery candidates (Loddenkemper and Kotagal, 2005). The aim of this article was to review literature data in order to better define this peculiar event during seizures and describe the clinical and electroencephalographic features of two new adult patients suffering from symptomatic temporal lobe epilepsy (TLE) with episodes of PIWD recorded by video-EEG.

## Methods

The electro-clinical characteristics in two adult patients are first described in detail. The review of the literature was performed on PubMed using the words: “peri-ictal drinking”, “peri-ictal water drinking”, “epilepsy/seizure” and “drinking/thirst”. To date, 51 cases with focal epilepsy and seizures associated with PIWD have been reported (Rémillard *et al.*,

1981; Despland *et al.*, 1985; Cascino and Sutula, 1989; Crapanzano *et al.*, 1993; Trinka *et al.*, 2003; Szucs *et al.*, 2007; Musilova *et al.*, 2010; Errguig *et al.*, 2013). Of these, seven cases reported by Janszky *et al.* (2007) were excluded from the analysis of data due to poor documentation (three patients with right-sided and four patients with left-sided TLE). For the same reason, 14 cases reported by Musilova *et al.* (2010) were excluded. The review was performed on 30 cases (table 1).

## Case studies

### Case 1

The first subject was a right-handed, 42-year-old male, suffering from recent onset of symptomatic focal epilepsy. At the last observation, he mainly presented two different seizure types: apparently generalized tonic-clonic (mostly during sleep) and sudden events of air hunger, facial flushing, and fear, without loss of consciousness. During these events, he was able to speak even if he appeared slightly disoriented. At the end of these episodes, he repetitively asked for water to drink. This latter aspect was so significant that when he experienced multiple seizures per day, he even developed water intoxication.

Ictal video-EEG showed rhythmic theta activity intermingled with spikes over the right temporal region, spreading over the right frontal and central regions, lasting for about 40 seconds. The episode was characterized by air hunger, facial flushing, and confusion in the first part; after 20 seconds, he started to drink water for the next two minutes (figure 1A). The semiology of seizures was similar to previous seizures, except that PIWD was now only sometimes reported. The patient was diagnosed as suffering from paraneoplastic limbic encephalitis involving bilateral temporal regions and the diencephalon (figure 2A). The primary cause was a testicular seminoma. It was not possible to rule out the presence of left temporal lobe seizures, however, during video-EEG monitoring, we recorded only right temporal-onset seizures.

### Case 2

The second subject was a right-handed, 76-year-old woman, affected by epileptic seizures since 1 year of age. Seizures were characterized by a sudden and brief cacosmia, sometimes preceded by sensation of epigastric constriction and followed by PIWD. A few months after the onset, she presented with status epilepticus (SE) characterized by confusion, incongruous behaviours, and repetitive water drinking requests; laboratory findings ruled out the presence of a dehydration state.

**Table 1.** Literature review of published cases.

| Reference                 | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology | Water<br>drinking | Aetiology        | Interictal<br>EEG                             | Ictal EEG                                                            | Neuro-<br>imaging<br>findings                            | Side | Treat-<br>ment | Follow-<br>up       |
|---------------------------|-------------|------------------------|------------------------------------|----------------------|-------------------|------------------|-----------------------------------------------|----------------------------------------------------------------------|----------------------------------------------------------|------|----------------|---------------------|
| Szucs<br>et al.<br>(2007) | 38/M        | NR                     | R-handed                           | NR                   | Ictal             | Symp-<br>tomatic | Inde-<br>pendent<br>bitemporal<br>spike focus | Right temporal<br>rhythmic theta<br>pocampal<br>sclerosis<br>(MRI)   | Right hip-<br>pocampal<br>sclerosis<br>(MRI)             | R    |                |                     |
|                           | 37/F        | NR                     | R-handed                           | NR                   | Postictal         | Symp-<br>tomatic | Right<br>temporal<br>sharp waves              | Right anterior<br>desynchroniza-<br>tion and<br>rhythmic<br>activity | Right tempo-<br>ral oligo-<br>astrocytoma<br>(MRI)       | R    |                |                     |
|                           | 36/F        | NR                     | R-handed                           | NR                   | Ictal             | Symp-<br>tomatic | Right<br>anterior<br>temporal<br>spikes       | Crescendo<br>right temporal<br>rhythmic<br>activity                  | Bilateral,<br>R>L hip-<br>pocampal<br>sclerosis<br>(MRI) | R    |                |                     |
|                           | 25/F        | NR                     | R-handed                           | NR                   | Ictal             | Crypto-<br>genic | Bitemporal<br>> left spikes                   | Left temporal<br>desynchroniza-<br>tion and<br>rhythmic<br>activity  | Normal<br>(MRI)                                          | L    | Surgery        | Engel<br>class I/II |
|                           | 38/M        | NR                     | R-handed                           | NR                   | Ictal             | Symp-<br>tomatic | Left anterior<br>temporal<br>spikes           | Left temporal<br>rhythmic<br>crescendo<br>pattern                    | Left hip-<br>pocampal<br>sclerosis<br>(MRI)              | L    |                |                     |
|                           | 34/F        | NR                     | R-handed                           | NR                   | Postictal         | Symp-<br>tomatic | Left<br>temporal<br>sharp waves               | Left temporal<br>theta rhythm                                        | Left hip-<br>pocampal<br>sclerosis<br>(MRI)              | L    |                |                     |

Table 1. (Continued)

| Reference                         | Age/<br>sex | Age/<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                                                                           | Water<br>drinking | Aetiology                                          | Interictal<br>EEG                        | Ictal EEG                                                                         | Neuro-<br>imaging<br>findings                | Side | Treat-<br>ment | Follow-<br>up |
|-----------------------------------|-------------|----------------------|------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|----------------------------------------------------|------------------------------------------|-----------------------------------------------------------------------------------|----------------------------------------------|------|----------------|---------------|
| Szucs<br><i>et al.</i><br>(2007)  | 30/M        | NR                   | R-handed                           | NR                                                                                                                                                             | Ictal             | Symp-<br>tomatic                                   | Bitemporal<br>independ-<br>ent<br>spikes | Left temporal<br>and frontal<br>desynchroniza-<br>tion and<br>crescendo<br>rhythm | Left hip-<br>pocampal<br>sclerosis<br>(MRI)  | L    |                |               |
|                                   | 30/F        | NR                   | R-handed                           | NR                                                                                                                                                             | NR                | Symp-<br>tomatic                                   | Left<br>temporal<br>spikes               | Left temporal<br>rhythmic<br>pattern                                              | Left<br>temporal<br>cavernoma<br>(MRI)       | L    |                |               |
| Trinka<br><i>et al.</i><br>(2003) | 41/M        | 5                    | L speech<br>(Wada test)            | Unresponsiveness,<br>right hand<br>automatism,<br>dystonic posturing<br>left upper<br>extremity                                                                | Postictal         | Symp-<br>tomatic<br>(Ammon's<br>horn<br>sclerosis) | Spikes (R/L<br>→ 60:40)                  | Bitemporal                                                                        | Right hip-<br>pocampal<br>sclerosis<br>(MRI) | R    |                | NR            |
|                                   | 52/F        | 50                   | L speech<br>(Wada test)            | Unresponsiveness,<br>oroalimentary and<br>right hand<br>automatizms,<br>dystonic posturing<br>left upper<br>extremity                                          | Postictal         | Symp-<br>tomatic<br>(gangli-<br>oglioma)           | Right spikes                             | Right temporal                                                                    | Right<br>temporal<br>tumour<br>(MRI)         | R    |                | NR            |
|                                   | 30/M        | 13                   | L speech<br>(Wada test)            | Motionless stare,<br>unresponsiveness,<br>oroalimentary and<br>right upper<br>extremity<br>automatism, ictal<br>water drinking,<br>secondary<br>generalization | Ictal             | Symp-<br>tomatic<br>(end-<br>folium<br>sclerosis)  | Right spikes                             | Right temporal                                                                    | Right hip-<br>pocampal<br>atrophy<br>(MRI)   | R    | Surgery        | NR            |

**Table 1.** (Continued)

| Reference                  | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                                                                               | Water<br>drinking | Aetiology                                            | Interictal<br>EEG   | Ictal EEG      | Neuro-<br>imaging<br>findings                | Side | Treat-<br>ment | Follow-<br>up |
|----------------------------|-------------|------------------------|------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|------------------------------------------------------|---------------------|----------------|----------------------------------------------|------|----------------|---------------|
| Trinka<br>et al.<br>(2003) | 43/F        | 8                      | L speech<br>(Wada test)            | Unresponsiveness,<br>oroalimentary and<br>right hand<br>automatisms,<br>dystonic posturing<br>left upper<br>extremity                                              | Postictal         | Symp-<br>tomatic<br>(Ammon's<br>horn<br>sclerosis)   | Right spikes        | Right temporal | Right hip-<br>pocampal<br>atrophy<br>(MRI)   | R    |                | NR            |
|                            | 42/M        | 13                     | L speech<br>(Wada test)            | Unresponsiveness<br>and right hand<br>automatisms                                                                                                                  | Postictal         | Symp-<br>tomatic<br>(Ammon's<br>horn<br>sclerosis)   | R/T (90:10)         | Right temporal | Right hip-<br>pocampal<br>sclerosis<br>(MRI) | R    |                | NR            |
|                            | 49/F        | 3                      | L speech<br>(Wada test)            | Unresponsiveness,<br>oroalimentary and<br>right hand<br>automatisms,<br>dystonic posturing<br>left upper<br>extremity                                              | Postictal         | Symp-<br>tomatic<br>(Ammon's<br>horn<br>sclerosis)   | Right spikes<br>(?) | Right temporal | Right hip-<br>pocampal<br>sclerosis<br>(MRI) | R    |                | NR            |
|                            | 36/F        | 32                     | L speech<br>(Wada test)            | Motionless stare,<br>unresponsiveness,<br>oroalimentary and<br>right hand<br>automatism, ictal<br>water drinking,<br>dystonic posturing<br>left upper<br>extremity | Ictal             | Symp-<br>tomatic<br>(fibrillary<br>astrocy-<br>toma) | Right spikes<br>(?) | Right temporal | Right fron-<br>totemporal<br>tumour<br>(MRI) | R    |                | NR            |

**Table 1.** (Continued)

| Reference                            | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance                                | Seizure<br>semiology                                                                                                                                                                | Water<br>drinking | Aetiology        | Interictal<br>EEG                                          | Ictal EEG                                                                             | Neuro-<br>imaging<br>findings                                                                               | Side                                     | Treat-<br>ment | Follow-<br>up |
|--------------------------------------|-------------|------------------------|-------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|------------------|------------------------------------------------------------|---------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------|------------------------------------------|----------------|---------------|
| Rémillard<br><i>et al.</i><br>(1981) | 33/M        | 2,5                    | R-handed                                                          | Confusion,<br>trembling, ictal<br>water drinking                                                                                                                                    | Ictal             | Symp-<br>tomatic | Bilateral<br>temporal<br>epileptic<br>activity<br>(R>L)    | EEG flattening<br>and rhythmic<br>spike<br>discharges<br>(L>R) (deep<br>electrodes)   | Enlargement (L)<br>of the left<br>lateral<br>ventricle,<br>particularly<br>of the<br>temporal<br>horn (PEG) | (L)                                      | NR             | NR            |
|                                      | 19/M        | 1                      | Ambidextrous                                                      | Staring,<br>disorientation,<br>head and eyes<br>right deviation,<br>salivation,<br>chewing,<br>hyperventilation,<br>papillary dilatation,<br>right upper<br>extremity<br>automatism | Pre-<br>postictal | NR               | Independent<br>bitemporal<br>epilepti-<br>form<br>activity | Fast rhythmic<br>activity starting<br>in the L<br>hippocampus<br>(deep<br>electrodes) | NR                                                                                                          | (L)                                      | NR             | NR            |
|                                      | 44/M        | 34                     | R-handed                                                          | Dizziness, nausea,<br>difficulty hearing,<br>"bars of an English<br>waltz"                                                                                                          | Ictal             | Symp-<br>tomatic | Right<br>temporal<br>focus                                 | NR                                                                                    | Diffuse<br>atrophy<br>with<br>asymmetry<br>(> right<br>frontal and<br>temporal<br>horns)<br>(PEG)           | (R)                                      | NR             | NR            |
|                                      | 21/F        | 10                     | L-handed<br>(preserved<br>ictal speech<br>during his<br>seizures) | Loss of contact,<br>looking around,<br>pronation and<br>retropulsion of<br>right arm                                                                                                | Ictal/postictal   | NR               | Left<br>temporal<br>epilepti-<br>form<br>activity          | Left frontal and<br>temporal<br>spikes                                                | NR                                                                                                          | (L- non-<br>dominant<br>hemi-<br>sphere) | NR             | NR            |

**Table 1.** (Continued)

| Reference                            | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                                                                       | Water<br>drinking | Aetiology        | Interictal<br>EEG                                                                              | Ictal EEG                                                                                                          | Neuro-<br>imaging<br>findings                                                                       | Side | Treat-<br>ment | Follow-<br>up |
|--------------------------------------|-------------|------------------------|------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|------------------|------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------|------|----------------|---------------|
| Rémillard<br><i>et al.</i><br>(1981) | 9/F         | 1,8                    | R-handed                           | Running about<br>asking for water,<br>confusion,<br>hyperventilation,<br>left limb stiffness                                                               | Ictal             | Symp-<br>tomatic | Right<br>temporal<br>focus                                                                     | Right temporal<br>high-voltage<br>sharp waves<br>with or without<br>slow waves                                     | Right hemi-<br>spherical<br>atrophy,<br>enlarged R<br>lateral<br>ventricle<br>(skull<br>radiograms) | (R)  | NR             | NR            |
|                                      | 24/F        | 15                     | L-handed                           | Feelings of not<br>being well,<br>trembling,<br>chewing,<br>swallowing,<br>lip-puckering,<br>flushing of the<br>face,<br>apprehension,<br>hyperventilation | Ictal             | NR               | Right infer-<br>omesial<br>temporal<br>area<br>rhythmic<br>theta<br>activity                   | Generalized<br>rhythmic theta<br>activity, some<br>right<br>frontotemporal<br>sharp waves<br>and delta<br>activity | NR                                                                                                  | (R)  | NR             | NR            |
|                                      | 50/F        | Ado-<br>lescent        | NR                                 | Odd sensation in<br>the head, water<br>drinking                                                                                                            | Pre?              | NR               | Right cen-<br>troparietal<br>dysfunction<br>without<br>clear<br>epilepto-<br>genic<br>activity | NR                                                                                                                 | NR                                                                                                  | (R)  | NR             | NR            |

Table 1. (Continued)

| Reference                            | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                          | Water<br>drinking | Aetiology        | Interictal<br>EEG                                                                                         | Ictal EEG                                               | Neuro-<br>imaging<br>findings                                                                   | Side | Treat-<br>ment | Follow-<br>up |
|--------------------------------------|-------------|------------------------|------------------------------------|---------------------------------------------------------------------------------------------------------------|-------------------|------------------|-----------------------------------------------------------------------------------------------------------|---------------------------------------------------------|-------------------------------------------------------------------------------------------------|------|----------------|---------------|
|                                      | 50/F        | 0,8                    | R-handed                           | Awakening, need<br>of water; GTCS;<br>heaviness and loss<br>of balance                                        | Ictal             | NR               | "disturbance<br>of cerebral<br>activity in<br>the left<br>temporal<br>and anterior<br>sylvian<br>regions" | NR                                                      | NR                                                                                              | (L)  | NR             | NR            |
| Rémillard<br><i>et al.</i><br>(1981) | 18/F        | 4                      | R-handed                           | Frightening,<br>swallowing,<br>staring, left arm<br>stiffness, head and<br>eyes left deviation<br>automatisms | Ictal             | Symp-<br>tomatic | Anterior<br>right<br>temporal<br>lobe epilep-<br>togenic<br>activity                                      | NR                                                      | Smaller left<br>hemicra-<br>nium with<br>enlarge-<br>ment of L<br>lateral<br>ventricle<br>(PEG) | (R)  | NR             | NR            |
|                                      | 53/F        | 5                      | R-handed                           | Dizziness,<br>swallowing, loss of<br>consciousness,<br>tonic-clonic<br>seizures                               | Ictal             | NR               | Anterior<br>and<br>midtempo-<br>ral right<br>temporal<br>lobe epilep-<br>togenic<br>activity              | Right anterior<br>and<br>midtemporal<br>epileptic focus | NR                                                                                              | (R)  | NR             | NR            |

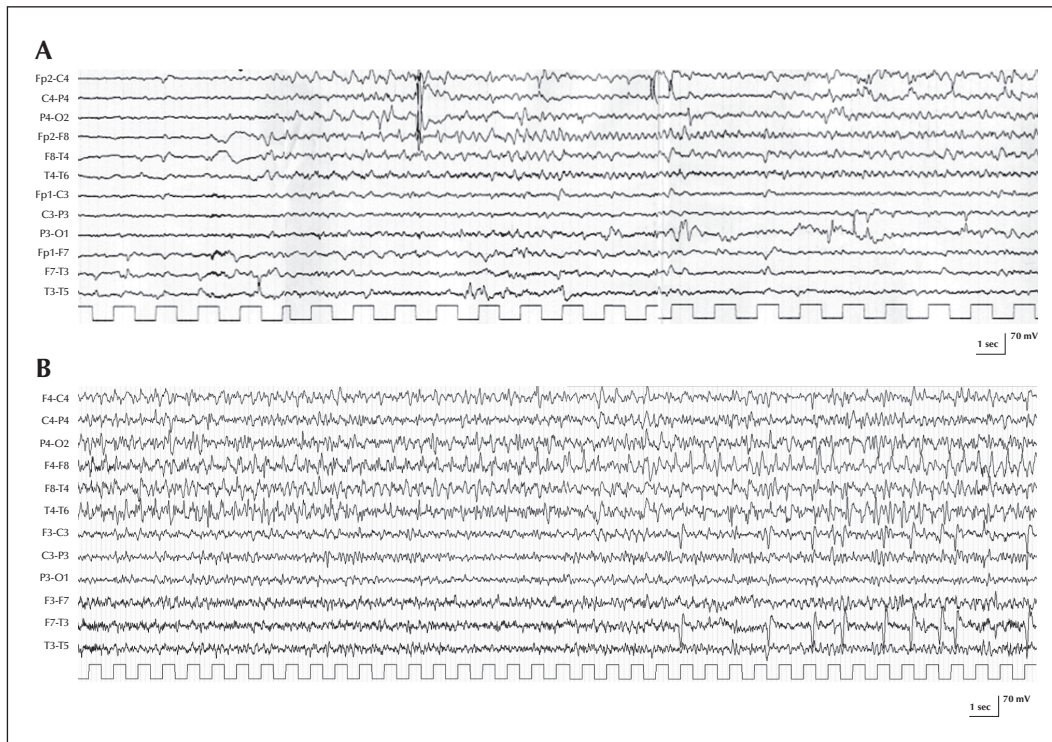
**Table 1.** (Continued)

| Reference                           | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                                                                       | Water<br>drinking | Aetiology                    | Interictal<br>EEG                                                            | Ictal EEG                                                                        | Neuro-<br>imaging<br>findings                                                                                                                                    | Side | Treat-<br>ment                    | Follow-<br>up                          |
|-------------------------------------|-------------|------------------------|------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|------------------------------|------------------------------------------------------------------------------|----------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------|------|-----------------------------------|----------------------------------------|
| Cascino<br>and Sutula<br>(1989)     | 39/F        | 11                     | R-handed                           | Motionless stare,<br>lip smacking, heat<br>turning to the left.<br>Lightheadedness<br>and palpitations,<br>urge to drink                                   | Ictal             | Symp-<br>tomatic             | Intermittent<br>epilepti-<br>form activity<br>in right<br>anterior<br>region | Right anterior<br>temporal<br>electrographic<br>seizure activity                 | Enlargement R<br>of the right<br>temporal<br>horn of the<br>lateral<br>ventricle<br>suggesting<br>atrophy of<br>the right<br>parahip-<br>pocampal<br>gyrus (MRI) | R    | Anterior<br>temporal<br>lobectomy | Seizure<br>freedom<br>since<br>surgery |
| Crapanzano<br><i>et al.</i> (1993)  | 35/F        | 24                     | R-handed                           | Staring,<br>disorientation,<br>drinking                                                                                                                    | Ictal             | Symp-<br>tomatic             | Right<br>temporal<br>sharp waves                                             | Right temporal<br>onset with<br>maximally<br>diffuse<br>high-amplitude<br>bursts | Right hip-<br>pocampal<br>atrophy<br>(MRI)                                                                                                                       | R    | NR                                | CBZ,<br>VPA                            |
| Despland<br><i>et al.</i><br>(1985) | 51/M        | 51                     | R-handed                           | Anxious behaviour,<br>chewing, weakness                                                                                                                    | Post              | Symp-<br>tomatic<br>(Glioma) | Right theta<br>rhythm                                                        | Right temporal<br>rhythmic<br>activity and<br>spikes                             | Right<br>fronto-<br>temporal<br>tumour                                                                                                                           | R    | NR                                | Death                                  |
| Errguig<br><i>et al.</i><br>(2013)  | 42/F        | 30                     | R-handed                           | Epigastric aura,<br>staring,<br>unresponsiveness,<br>oro-alimentary<br>automatisms,<br>dystonic posturing<br>of the left hand,<br>ictal water<br>drinking. | Ictal             | Symp-<br>tomatic             | Epileptic<br>discharges<br>in the right<br>anterior<br>temporal<br>lobe      | Right anterior<br>temporal<br>electrographic<br>seizure activity                 | Right hip-<br>pocampal<br>sclerosis<br>(MRI)                                                                                                                     | R    | Surgery                           | Engel I                                |

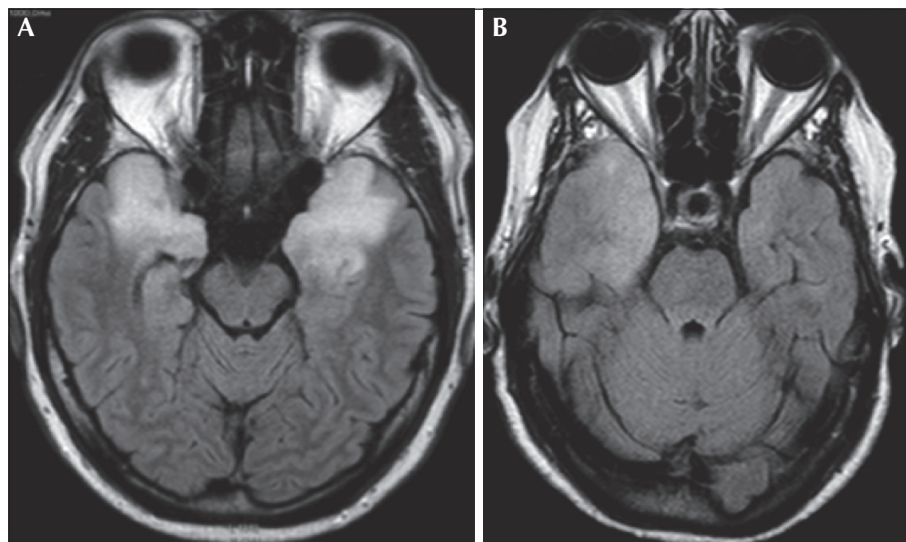
**Table 1.** (Continued)

| Reference                          | Age/<br>sex | Age at<br>onset<br>(y) | Handedness/<br>speech<br>dominance | Seizure<br>semiology                                                                                                                    | Water<br>drinking | Aetiology        | Interictal<br>EEG                                                       | Ictal EEG                                                                                     | Neuro-<br>imaging<br>findings                                                                                                            | Side | Treat-<br>ment               | Follow-<br>up      |
|------------------------------------|-------------|------------------------|------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------|-------------------|------------------|-------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------|------|------------------------------|--------------------|
| Errguig<br><i>et al.</i><br>(2013) | 36/F        | 8                      | R-handed                           | Epigastric aura,<br>tachycardia,<br>staring,<br>hypersalivation,<br>motor and verbal<br>automatisms with<br>feeling of thirst.          | Ictal             | Symp-<br>tomatic | Epileptic<br>discharges<br>in the right<br>anterior<br>temporal<br>lobe | Right anterior<br>temporal<br>electrographic<br>seizure activity                              | Right hip-<br>pocampal<br>sclerosis<br>(MRI)                                                                                             | R    | Awaiting<br>surgery          | NR                 |
| Case 1<br>(this<br>study)          | 42/M        | 42                     | R-handed                           | Air hunger, facial<br>flushing, fear,<br>without loss of<br>consciousness,<br>able to speak, he<br>repetitively asked<br>water to drink | Ictal             | Symp-<br>tomatic | Bilateral<br>temporal<br>epileptic<br>activity                          | Rhythmic<br>theta activity<br>intermingled<br>with spikes<br>over right<br>temporal<br>region | Paran-<br>eoplastic<br>limbic<br>encephalitis<br>involving<br>bitemporal<br>regions and<br>dien-<br>cephalon<br>(testicular<br>seminoma) | ?    | Primary<br>tumour<br>surgery | Seizure<br>freedom |
| Case 2<br>(this<br>study)          | 76/F        | 75                     | R-handed                           | SE characterized by<br>confusion,<br>incongruous<br>behaviours, and<br>repetitive WD<br>requests                                        | Ictal             | Symp-<br>tomatic | Right<br>temporal<br>theta<br>rhythm                                    | Rhythmic<br>spikes over<br>right temporal<br>region                                           | Right<br>temporal<br>glioblas-<br>toma                                                                                                   | R    | i.v. AEDs                    | Death              |

M: male; F: female; y: year; NR: not reported; R: right; L: left; CBZ: carbamazepine; VPA: valproic acid; SE: status epilepticus; iv: intravenous; AEDs: antiepileptic drugs. (R) or (L): data obtained indirectly; see text for details.



**Figure 1.** Ictal EEG of both patients. (A) Case 1 with limbic encephalitis. EEG shows a low-voltage fast activity over the right temporal region, followed by rhythmic spikes in the same region. (B) Case 2 with right temporal lobe glioblastoma. EEG shows rhythmic spikes over the right temporal and frontal regions.



**Figure 2.** Brain MRI of both patients. (A) Case 1: FLAIR sequences showing bilateral temporal lobe hyperintensity, suggestive of limbic encephalitis. (B) Case 2: FLAIR sequences showing right temporal lobe hyperintensity.

She immediately underwent video-EEG monitoring which revealed rhythmic spikes over the right temporal region (*figure 1B*). Status persisted despite the use of i.v. antiepileptic treatment. Brain MRI, performed before SE, showed the presence of a right temporal glioblastoma (*figure 2B*). She died due to complications during the hospitalisation.

## Review of previous reported cases and discussion

PIWD is a stereotyped behaviour not always recognized as an epileptic automatism. It can be observed as an ictal or postictal sign, and rarely it represents *per se* the clinical manifestation of an ictal

temporal discharge. In our patients, it was part of symptomatic TLE.

Lennox and Cobb (1933) firstly reported the urgent need to drink water as part of an epileptic seizure. In their review on aura in epilepsy, three patients asked for water for an undetermined reason, and three had a sensation of dryness of the mouth.

Based on an extensive review of patients with autonomic signs associated with TLE, PIWD was reported in 14.4% of patients (Musilova *et al.*, 2010). Janszky *et al.* (2007) observed PIWD in 7% of patients, Szucs *et al.* (2007) in 14.5%, and Trinka *et al.* (2003) in 15.3%. In these cases, drinking was encountered in patients with TLE in the context of focal symptomatic epilepsy.

Table 1 summarizes the main clinical findings of all reported cases (Rémillard *et al.*, 1981; Despland *et al.*, 1985; Cascino and Sutula, 1989; Crapanzano *et al.*, 1993; Trinka *et al.*, 2003; Szucs *et al.*, 2007; Errguig *et al.*, 2013). All patients presented with TLE, and all but one had symptomatic epilepsy. Twenty-four of 32 cases (including our cases) had involvement of the right non-dominant hemisphere, and eight of the left hemisphere.

Twenty-one patients were reported to have ictal water drinking (WD); eight had postictal WD and two had ictal/postictal WD. Both our patients had ictal WD. Only in a few cases was the timing of PIWD reported to correlate with the EEG; in cases reported by Errguig *et al.* (2013), the onset of fast rhythms in the right anterior temporal lobe was coincident with sensation of thirst. In cases with postictal WD, it remains unclear whether this finding might reflect a kind of inhibition or release phenomenon of the network involved in thirst.

Nineteen patients had brain MRI and four had pneumoencephalography or analogue X-ray of the skull. Neuroimaging was not available in the other cases. Aetiology was hippocampal sclerosis in 14 cases, and temporal tumour in five cases. Both of our cases had brain MRI, and aetiology was paraneoplastic limbic encephalitis in one and glioblastoma in the other.

The lateralizing value of PIWD is a point of debate. Although there are several articles in the literature proposing that it might represent a lateralizing symptom in TLE, there is no sufficient evidence to confirm this hypothesis. It was firstly proposed that the presence of PIWD represents a clear sign of involvement of the non-dominant temporal lobe (Trinka *et al.*, 2003; Musilova *et al.*, 2010), but this result was not confirmed by successive studies (Rémillard *et al.*, 1981; Janszky *et al.*, 2007; Szucs *et al.*, 2007).

The relatively small number of patients and seizures analysed, as well as the small number of patients investigated with stereo-EEG, are the two major issues to be highlighted when referring to the lateralizing value of PIWD.

Patients reported by Trinka *et al.* (2003) had a right (non-dominant) TLE, and this was documented through Wada testing performed for all of them. In the other published cases, the side of the temporal epileptogenic region was variable. Szucs *et al.* (2007) reported eight of 55 adult patients presenting with PIWD in the context of TLE; five had left and three right TLE, therefore the lateralization value of PIWD could not be confirmed.

In cases reported by Rémillard *et al.* (1981), the authors did not refer to any lateralizing value. However, 6 of 10 patients had epileptiform discharges on the right hemisphere. The other reported cases (Despland *et al.*, 1985; Cascino and Sutula, 1989; Crapanzano *et al.*, 1993; Errguig *et al.*, 2013) had symptomatic TLE with involvement of the right non-dominant hemisphere. In our patients, one had bilateral TLE and the other right TLE, therefore no clear conclusion could be drawn.

Overall, outcome is good; seizure freedom was reported in 7 of 10 patients presenting with PIWD after resective surgery (Wieser's class I) (Trinka *et al.*, 2003) and a favourable post-operative outcome was also achieved (Engel class I or II) in patients reported by Szucs *et al.* (2007). Two other patients were reported to be seizure-free after surgery (Cascino and Sutula, 1989; Errguig *et al.*, 2013). In addition, one patient was reported to be under AED treatment (Crapanzano *et al.*, 1993), one died (Despland *et al.*, 1985), and another was awaiting surgical treatment (Errguig *et al.*, 2013).

One of our patients died due to complications during the hospitalization and the other became seizure-free after surgery for testicular seminoma with remission of paraneoplastic limbic encephalitis.

Regarding the pathophysiology, it has been hypothesized that, in the context of a TLE, the propagation of mesial temporal epileptiform discharges via pathways between the hypothalamus and the mesial temporal structures might cause thirst, activating water seeking (Trinka *et al.*, 2003). It was suggested that the lateralizing value of PIWD could be explained by asymmetric representation of the central autonomic network responsible for fluid control, thirst, and water-seeking behaviour (Denton *et al.*, 1999).

The mechanism underlying PIWD was reported to be electrographically associated with seizures starting in the amygdala, hippocampus, and parahippocampal gyrus (Rémillard *et al.*, 1981), but it can also be induced in rats by stimulation of the lateral hypothalamus (Mogenson and Stevenson, 1961). The hypothalamus is linked to limbic structures by a variety of multi-synaptic pathways (Brodal, 1981). Efferent pathways from the hippocampus and amygdala that project to the hypothalamus have also been implicated in water balance and drinking behaviour (Martin and Reichlin, 1987).

Like ictal fear, ictal drinking may be the expression of epileptic activation of a behavioural mechanism integrated in temporo-limbic structures, which are related to some fundamental drive.

Seizure semiology of most of these patients was characterized by staring, sometimes preceded by aura, and followed by oral or motor automatisms, but motor phenomena were also reported (see *table 1*). Our cases presented with episodes of temporal aura, confusion or loss of contact, and then water request. Patients with TLE may react to an unpleasant feeling in the mouth or the feeling of thirst, resulting in a desire to drink.

We believe that PIWD, as part of an epileptic seizure, might be considered a rare automatic behaviour, like other automatisms frequently reported in epileptic seizures. Automatisms are more frequently described in patients with TLE, and involvement of the temporal lobe in most of the published cases might provide an explanation for this. On the other hand, complex motor behaviours require coordination of the motor nuclei of the brainstem, and an involvement of such structures cannot be ruled out. Hypothalamic activation, also, has been hypothesized as an explanation for drinking in the peri-ictal period (Trinka et al., 2003). It is possible that drinking within epileptic seizures is a release phenomenon rather than activation of distinct symptomatogenic cortex. Whether this rare event is the result of activation of particular areas of cortex or a release phenomenon is not clear. □

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