

A Young man with Peri-ictal water drinking

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ABSTRACT

تم ربط السمات العصبية بالصرع الناشئ من الفص الصدغي والتي يمكن رؤيتها أثناء حدوث التشنجات وتلعب دوراً مهماً في تحديد الجانب البؤري المصاب ومن بين السمات المعروفة والنادر حدوثها هي شرب الماء أثناء نوبات التشنجات. هنا نقدم حالة رجل يبلغ من العمر 31 عاماً مصاباً بالصرع الجزئي الناشئ من الفص الصدغي المقاوم للأدوية ويصاحبه نوبات شرب الماء. وقد تم إعتبار المريض مرشحاً لجراحة الإستئصال الجزئي بعد عمل الفحوصات اللازمة التي شملت التخطيط الكهربائي للدماغ المراقب بالفيديو والرنين المغناطيسي للدماغ وتقييم الذاكرة بواسطة علم النفس العصبي التي أشارت إلى تحديد البؤرة الصرعية في الفص الصدغي من الجانب غير المهيمن من الدماغ وكان للمريض نتيجة ممتازة بعد استئصال الفص الصدغي الأمامي من الجانب الأيمن (غير المهيمن). إلى يومنا هذا لم يتم تحديد الأهمية للتخصيص الجانبي لشرب الماء أثناء التشنجات بشكل نهائي ويرجع ذلك إلى العدد المحدود من الحالات المنشورة. تسلط هذه الحالة الضوء على العلاقة التي لوحظت سابقاً بين شرب الماء أثناء التشنجات وربطها بالجانب غير المهيمن من الدماغ في مرضى الصرع.

Neuro-vegetative features have been linked to epilepsy arising from the temporal lobe, which can be seen during ictal events and play an important role in determining the focal side of the lesion. Among the rare known features is peri-ictal water drinking (PIWD). Here, we present the case of a 31-year-old male with refractory temporal epilepsy, having episodes of PIWD. The patient was considered a candidate for resective surgery and was investigated accordingly, including long-term video electroencephalogram (EEG), brain magnetic resonance imaging (MRI), and neuropsychology assessment, which pointed towards a lesioned temporal lobe over the non-dominant hemisphere. The patient had an excellent outcome following right anterior temporal lobectomy. The lateralization significance of PIWD has not yet been established definitively in the literature, partly due to a limited number of published cases. This case highlights the previously observed association between PIWD and the involvement of the non-dominant hemisphere in epilepsy patients.

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The initial description of peri-ictal water drinking (PIWD) dates back to the early twentieth century with observations by William G. Lennox and Stanley Cobb.¹ However, drinking water is a usual habitual action, so it is difficult for patients or witnesses to consider this behavior as part of ictal manifestation, which made PIWD largely underestimated. Neuro-vegetative features such as mydriasis, transient hypo- or hypertension, and an increase in heart rate are well known and occur quite frequently in temporal lobe seizures. Many other features are described and occur much less frequently, as is the case in PIWD.² Review of the literature showed only a handful of studies looking into PIWD, and no definite evidence exists to answer all questions about its relevance to temporal lobe epilepsy (TLE); however, a possible lateralizing value has been hypothesized.³ We present a case of right symptomatic TLE secondary to mesial temporal sclerosis (MTS) with PIWD behavior episodes confirmed by ictal video-electroencephalogram (EEG) that was recorded in an epilepsy monitoring unit. We also reviewed the literature to evaluate the localization and lateralization significance of this manifestation in TLE.

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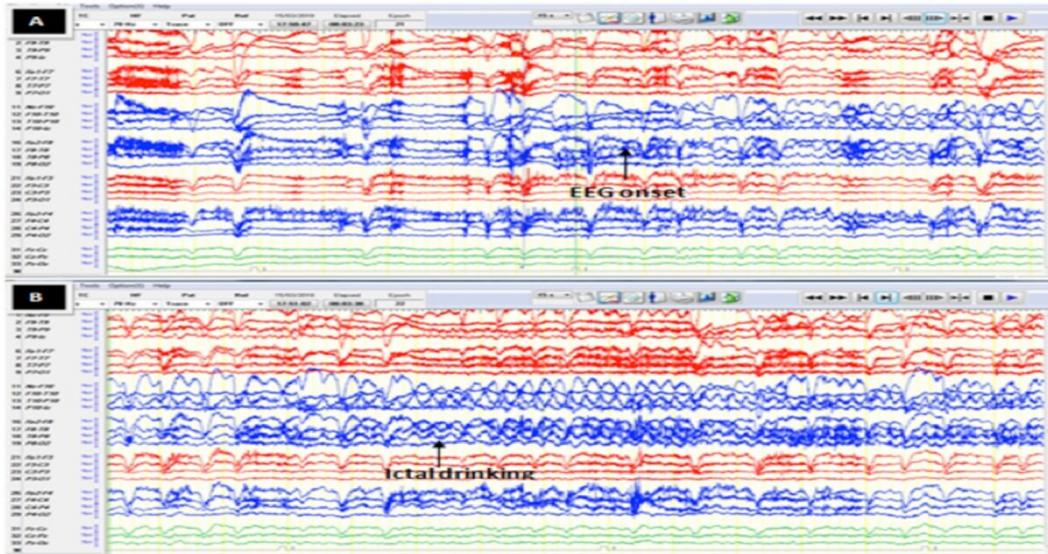


Figure 1 - Electroencephalogram (EEG) (A) Ictal EEG onset with rhythmic discharges started from the right temporal region; (B) High amplitude rhythmic discharges in the right temporal region (along with the beginning of ictal water drinking 30 seconds after EEG onset).

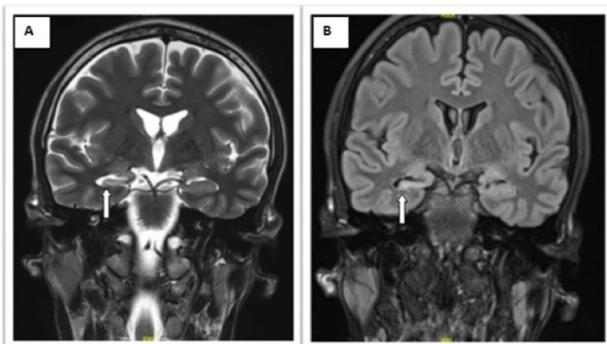


Figure 2 - The coronal T2WI (A) and FLAIR (B) images showing right-sided mesial temporal sclerosis. Notice the increased signal and small hippocampus with loss of normal internal architecture of the right hippocampus and dilated right temporal horn.

Case Report. Patient information and clinical findings. A 31-year-old left-handed male known to have epilepsy since the age of 7 years presented to our epilepsy center at King Fahad Medical City, Riyadh, Kingdom of Saudi Arabia. His seizures were characterized by palpitations and thirst followed by right-hand automatism. He reported partial intact awareness and orientation at first, but later he could not recall what had happened. No other seizure types or history of febrile convulsion were documented. The treatment regimen consisted of valproic acid and levetiracetam (1 g bid for each drug) with no seizure control. Clinical and neurological examinations were unremarkable.

Diagnostic assessment. The patient was admitted to our epilepsy center for preoperative evaluation, including video-EEG monitoring, brain magnetic resonance imaging (MRI), positron emission tomography (PET) scanning, and a neuropsychological assessment. The patient had 6 seizures during hospitalization secondary to the gradual discontinuation of antiepileptic drugs (AEDs). The episodes were characterized by sudden mild headaches, palpitations, and excessive drinking of water followed by left-hand dystonic contraction. During these episodes, he was able to speak and gave meaningful answers to questions; later, he became disoriented. Ictal EEG changes began 30 sec before clinical onset and showed a buildup of rhythmic theta activity over the right anterior and mid-temporal head regions (Figure 1A, B). No EEG changes were noted post-ictally. Brain MRI showed right mesial temporal sclerosis (Figure 2A, B), and a PET scan revealed hypometabolism in the right temporal lobe. Neuropsychological evaluation results were within normal limits.

Therapeutic intervention, follow-up, and outcome. The patient underwent a right anterior temporal resection. He was seizure-free at the one-year follow-up; however, he remained on AEDs. His follow-up EEG showed residual epileptic activity (Figure 3).

Discussion. The PIWD is an uncommon feature described in patients with lesional and non-lesional epilepsy of the temporal lobe. It is defined as a stereotyped action seen across all age groups during

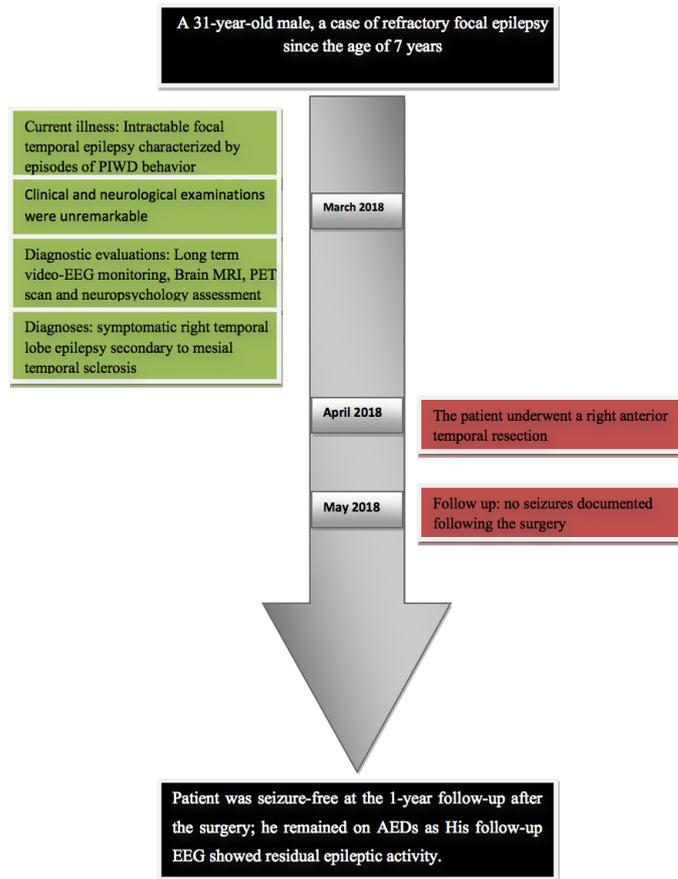


Figure 3 - Time line figure of the presented case.

seizures or within 1 to 2 minutes of their onset.³ The described case in this report concerned lesional epilepsy of the temporal lobe as documented in neuroimaging over the right side with automatic PIWD during video-EEG documented seizures, confirming the ictal nature of this manifestation.

To date, 64 cases of epileptic seizures associated with PIWD have been reported.⁴ The rate of reported cases of PIWD seen in TLE ranged from 7% to 15.3%.^{2,3,5,6} It was observed in 15.3% of 386 seizures in 65 patients with focal epilepsy reported by Trinka et al.³ In a study of 55 adult patients with TLE, PIWD was seen in 8 patients (14.5%).⁶ Janszky et al⁵ observed PIWD in 7% of 141 adult patients with TLE. Musilová et al² reported PIWD in 14.4% of 380 seizures in 97 patients with TLE-associated vegetative signs.

Generally, favorable surgical outcomes have been described for TLE patients with PIWD. A case series published by Trinka et al.³ demonstrated a complete response to surgical treatment in 70% of the studied

cohort (Wieser's class I). Similar favorable results of surgery were seen with a group of patients studied by Szűcs et al.⁶ (Engel Class 1 or 2) with the outcome maintained for a few years of postoperative follow-up. Our patient was seizure-free at the one-year follow-up after surgery.

Normal drinking behavior is controlled by vascular volume and tissue osmolality. Information from peripheral receptors is processed in the hypothalamus and then conveyed to various brain structures that initiate a search for water and drink.⁷ Furthermore, the hippocampus and the amygdala are linked to the hypothalamus through several multi-synaptic pathways that are involved in the regulation of water balance and drinking behavior.⁸ Water-seeking behavior is triggered by the involvement of the hypothalamus, which is affected by abnormal electrical activity generated by the temporal lobe.⁹

The PIWD is thought to reflect involvement of the non-dominant hemisphere because neural networks

involved in fluid balance, thirst, and water-seeking are normally asymmetrical and are likely more active in the non-dominant side.¹⁰ The lateralizing value of PIWD remains incompletely determined. Some studies have linked PIWD to significant lateralization to the non-dominant hemisphere in patients with TLE.^{2,3} Other reports could not demonstrate clear evidence for the lateralization value of PIWD.^{5,6} This uncertainty could be due to the rapid spread of epileptic discharges from the focal source of the seizures to the contralateral side of the temporal lobe, or it may be due simply to the few reported cases of PIWD.⁶

In conclusion, PIWD is a rare and important vegetative symptom of TLE that can be a valuable lateralizing sign to the non-dominant hemisphere. We report here a case of PIWD as a manifestation of TLE in the non-dominant hemisphere; however, the association of PIWD with a non-dominant hemisphere has not yet been definitively established in the literature. Given the significance of PIWD in planning surgical resection, a more comprehensive and powerful study with enrollment of more patients is needed. Furthermore, the lateralization could be supported and confirmed by invasive EEG recordings.

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Case Reports

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