

# Hot water epilepsy: Phenotype and single photon emission computed tomography observations

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## Abstract

We studied the anatomical correlates of reflex hot water epilepsy (HWE) using multimodality investigations viz. magnetic resonance imaging (MRI), electroencephalography (EEG), and single photon emission computed tomography (SPECT). Five men (mean age:  $27.0 \pm 5.8$  years) with HWE were subjected to MRI of brain, video-EEG studies, and SPECT scan. These were correlated with phenotypic presentations. Seizures could be precipitated in three patients with pouring of hot water over the head and semiology of seizures was suggestive of temporal lobe epilepsy. Ictal SPECT showed hyperperfusion in: left medial temporal — one, left lateral temporal — one, and right parietal — one. Interictal SPECT was normal in all five patients and did not help in localization. MRI and interictal EEG was normal in all the patients. The clinical and SPECT studies suggested temporal lobe as the seizure onset zone in some of the patients with HWE.

## Key Words

Hot water epilepsy, magnetic resonance imaging, single photon emission computed tomography, temporal lobe epilepsy

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## Introduction

Seizures precipitated by a specific sensory stimulus are described as reflex epilepsy. Reflex epilepsies are interesting, not only due to their being intriguing neurobehavioral phenomenon, but also serves as a model and provides insight into the pathophysiology and understanding of epilepsy in general. Hot water epilepsy (HWE), reflex epilepsy, is precipitated by the stimulus of bathing in hot water poured over the head.<sup>[1-5]</sup> The diagnostic scheme proposed by the International League against Epilepsy (ILAE) in 2001 included HWE, as a type of reflex epilepsy.<sup>[6,7]</sup>

The exact etiopathogenesis of this type of epilepsy is not clear, but several factors including genetic factors, environmental factors, consanguineous marriages, and habit of taking bath with high temperature water have been postulated as probable reasons.<sup>[8]</sup> Previous studies have reported that complex partial seizure (CPS) mainly of temporal lobe origin occurred in 67-100%

of the patients.<sup>[2,4,5]</sup> Interictal electroencephalography (EEG) is usually normal in most of the cases, but few case studies had shown localized temporal lobe discharges.<sup>[2,3,9]</sup> However, brain magnetic resonance imaging (MRI) does not show any structural abnormality except for isolated case reports of associated hippocampal sclerosis.<sup>[3,9]</sup> There is paucity of studies regarding single photon emission computed tomography (SPECT) and HWE especially with its clinical and EEG correlation.

The aim of this study was to study the regions of the brain involved in patients with HWE by SPECT, and video-EEG observations.

## Patients and Methods

Five patients with HWE were evaluated prospectively at a university teaching hospital, a major tertiary care referral center for neuropsychiatric disorders in south India. The diagnosis of HWE was based on the Commission of the ILAE.<sup>[6]</sup> The recruitment and evaluation was carried out under the supervision of neurologists with special interest in epilepsy research (PSC/SS). Approval from the Institute Ethics Committee and informed written consent was obtained from patients or relatives. All the patients underwent detailed evaluation with clinical history, physical and neurological examination, and investigations with SPECT ( $n = 5$ ) and four patients underwent MRI of brain on a 3T MRI machine (Achieva, Philips) with standard epilepsy protocol.

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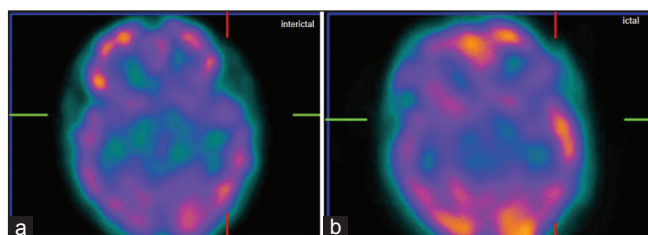
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Recruited patients were evaluated and data related to detailed history of epilepsy including bathing habits, precipitating factor — hot water bath, triggering areas, seizure semiology from eye witness, age at onset of seizures, frequency of seizures, history of epilepsy/HWE in the family, previous history of febrile convulsions/birth related injuries, gross development; personal/family medical histories were recorded. Clinical videography using mobile phone/video camera was performed for documentation of the seizures in patients with hot water head bath. Scalp EEGs were recorded on 16-channel “Galileo NT (EBN)” machine, employing international 10-20 system of electrode placement using standard parameters and procedures, for example, high filter-70 Hz; low filter-0.1 Hz; recording time: 30 min; sensitivity: 7 µV/mm; sampling rate: 256 Hz.

Single photon emission computed tomography scans were performed on Symbia T6 True Point SPECT machine (Siemens, Erlangen, Germany). Radiotracer used in this study was 99mTc-ethylene cysteinate dimer (ECD). During ictal and interictal studies, 20 mCi of freshly prepared 99mTc-ECD was injected intravenously followed by normal saline flush. Images of ictal and interictal studies were acquired, between 45 min and 1 h after radioisotope injection, using a dual-head gamma camera equipped with low energy high resolution collimators. All the patients underwent an identical protocol. Interictal SPECT was carried out in all the patients with seizure free interval of 24 h. Ictal SPECT was performed after inducing seizures with hot water bath and radiotracer (ECD) was injected immediately at the onset of the seizures. Tracer was injected with mean interval of 8 s after onset of ictus with range of 5-12 s from the onset of clinical seizure. Video recording of ictal event was done in all patients during procedure. Interictal and ictal SPECT images



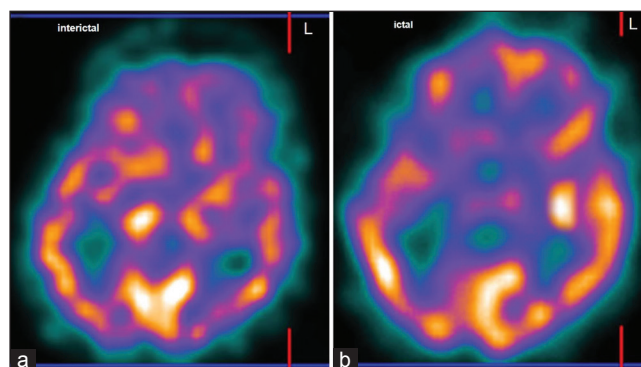
**Figure 1: Single photon emission computed tomography imaging in a patient (no. 2) with hot water epilepsy showing normal interictal scan (a) and hyperperfusion of left lateral temporal region in the ictal scan (b)**

were evaluated for evidence of focal hyper- or hypo-perfusion abnormality by visual analysis by nuclear medicine specialist.

**Results**

The age ranged from 17 to 32 years (mean: 27 ± 5.8 years; median: 28 years) in five patients with HWE. The mean age at the onset was 21.8 ± 4.7 years. Two patients had family history of HWE with the recessive inheritance pattern. There was no neurological deficit. One patient had a history of febrile seizures in childhood. One patient in addition had spontaneous unprovoked seizures. Structural brain imaging was normal in all. All patients had normal interictal EEG. All had a well-controlled seizure while on intermittent prophylactic clobazam about 60-90 min before hot water bath except one with spontaneous seizure who required continuous treatment with additional carbamazepine.

Three attacks could be precipitated with hot water. All the three attacks were during middle of bath with pouring of hot water over head. There was dazed look, irrelevant speech, and lethargy lasting 20 s to 2 min followed by postictal drowsiness. One patient required higher temperature (48°C) to precipitate attack. Others required hot water of about 40°C. Patients with attacks precipitated by HWE underwent ictal SPECT which showed hyperperfusion in left lateral temporal — one, left medial temporal — one, and right parietal — one [Figures 1 and 2]. However, there was delay (beyond 15 s) in injecting 99mTc-



**Figure 2: Single photon emission computed tomography imaging in a patient (no. 1) with hot water epilepsy showing normal interictal scan (a) and hyperperfusion of left medial temporal and slightly on right lateral temporal regions in the ictal scan (b)**

**Table 1: Localization in HWE**

Case no.	Semiology	Clinical attack localization	Interictal EEG	Ictal SPECT localization (n = 3)*	Imaging of brain	Final localization^
1	Dazed look, staring of eyes, behavioral arrest, unresponsive	Temporal	Normal	Ictal: Left medial temporal	Normal MRI	Left medial temporal
2	Dazed look, staring of eyes, behavioral arrest, unresponsive	Temporal	Normal	Ictal: Left lateral temporal	Normal MRI	Left lateral temporal
3	Dazed look, staring of eyes, behavioral arrest, irrelevant speech	Temporal	Normal	Ictal: Normal	Normal MRI	Temporal ^
4	Light headedness, behavioral arrest, irrelevant speech	Temporal	Normal	No attacks could be recorded for SPECT	Normal MRI	Temporal
5	Dazed look, staring of eyes, behavioral arrest, unresponsive	Temporal	Normal	No attacks could be recorded for SPECT	Normal CT	Temporal

\*Interictal SPECT: normal in all; based on clinical semiology. EEG = Electroencephalograph, SPECT = Single photon emission computed tomography, MRI = Magnetic resonance imaging, CT = Computed tomography, HWE = Hot water epilepsy; ^Final localization is temporal based on clinical findings and the tests they underwent

ECD in the last patient. Interictal SPECT was normal in all five patients and did not help in localization. The details are provided in Table 1.

Clinically, all had temporal lobe seizures with ictal SPECT clearly supporting it in two patients. The interictal EEG and SPECT studies were normal in all of them. Structural brain imaging was normal in all the patients.

## Discussion

All the five patients had clinical semiology indicative of CPS with onset from temporal lobe. MRI was normal in all five patients in accordance with earlier studies. However, there are isolated case reports of mesial temporal sclerosis (MTS) associated with HWE.<sup>[3,9]</sup> Interictal EEG was normal in all five cases. There are only few reports of temporal discharges on EEG.<sup>[10]</sup> Similarly, in a previous report of EEG changes in 70 patients with HWE, 15 had interictal EEG abnormalities in the frontotemporal region.<sup>[9]</sup> In our study, ictal SPECT in two cases showed medial and lateral temporal lobe hyperperfusion. In a case report by Lisovoski *et al.* (1992), temporal lobe hyperperfusion in ictal SPECT was noted in a patient with HWE.<sup>[11]</sup>

Animal models have suggested aberrant thermoregulatory system sensitive to rapid rise in temperature causing hyperthermic kindling.<sup>[11]</sup> Recent study in 20 patients with HWE has shown parasympathetic dysfunction supporting role of autonomic system in HWE.<sup>[12]</sup> Our patient had seizures with hot water only and one of the patient required higher temperature of water. This finding suggested a role of temperature rise in triggering seizure.<sup>[11]</sup> Temporal lobe is part of limbic system, which regulates temperature and autonomic function through hypothalamus. There might be genetic predisposition based previous studies, but how this leads to temporal lobe neuronal hyperexcitability with hot water bath is not yet known. It might as well be concluded that dysregulation of hypothalamus and limbic system in HWE with genetic susceptibility.

This clinical and SPECT studies reiterates temporal lobe as the seizure onset zone in some of the patient with HWE. Further,

reports of EEG changes, rare occurrence of MTS, and animal model studies point to temporal lobe involvement.

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