

Case Report

Hot water epilepsy with cerebral lesion: A report of five cases with cranial MRI findings

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Abstract

Hot water epilepsy (HWE) is included in the reflex epilepsies. Although, in general, not common, HWE is concentrated in certain regions of the world. Different bathing habits and genetic factors may be responsible for the high incidence of HWE in these regions. However, the exact pathogenesis of HWE is not known. The facts that complex partial seizures are the most common clinical presentation and EEG recordings show an epileptic focus in the temporal lobe suggested the presence of a structural lesion in the temporal lobe. To our knowledge, however, there were no demonstrable structural changes on MRI and CT scans except in a few case reports. Here, we describe an additional five cases of HWE having an intracranial pathology, for example, hippocampal sclerosis, dysplasia, and a huge cystic lesion. We believe that investigations with new detailed neuroimaging techniques, in addition to experimental and clinical studies, might help us to understand the mechanism of this reflex epilepsy.

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1. Introduction

Reflex seizures are precipitated by a specific sensory stimulus such as reading, eating, flickering light and color patterns, auditory stimulation, and hot water immersion in some epileptic patients. Reflex epilepsy due to bathing in hot water has been named “hot water” or “bathing” epilepsy [1–8]. In addition to case reports [3,7,9,10], small or large series of patients with hot water epilepsy (HWE) [2,3,6,8] have also been reported. Most of the series reported were from southern India and Turkey. In these regions, the percentages of patients with HWE among all patients with epilepsy were 6.9 and 0.6%, respectively. The reasons for the high incidence in these regions were speculated to be

bathing habits, like the pouring of hot water over the head, and genetic factors [1,2].

The exact pathogenesis of HWE is not known, but interestingly, complex partial seizures are common in this type of epilepsy [1–3]. Ictal and interictal electroencephalographic recordings of patients with HWE reveal focal paroxysmal activity in the temporal or frontal lobes [2,7,10–12]. It was suggested that a lesion in the temporal lobe, which could not be shown by neuroradiologic investigations, might be responsible for HWE [10]. However, only three case reports of HWE with structural cerebral lesions demonstrated by computed tomography (CT) or magnetic resonance imaging (MRI) have been reported so far [12–14]. Herein we describe an additional five cases of HWE with cerebral lesions from three different centers in Turkey. These patients were included in a multicenter study on HWE in Turkey conducted by a national HWE study group in which the authors participated. According to this study, 5 of 89 patients with HWE had cerebral lesions [15].

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2. Case reports

2.1. Patient 1

A 17-year-old boy was admitted to the emergency room after having a generalized tonic–clonic seizure during sleep. He awakened with a feeling of nausea and urinary urgency before the seizure. His physical and neurological examinations were unremarkable. A CT scan of the head was also normal. The patient denied a previous history of seizures. It was, however, noted that since the age of 7, he had experienced a few minutes of nausea while bathing with hot water. These attacks persisted after he left the bathroom. He bathed by pouring water over his head in the shower. These episodes did not occur during short bathing sessions using warm water. He also stated that he started having these episodes of nausea after awakening once a month at the age of 12. The attacks lasted a few minutes and were followed by fatigue and weakness. None of them were associated with automatism, clonic movements, or myoclonic jerks. He had not experienced loss of consciousness except during the last generalized tonic–clonic seizure.

There was no history of febrile convulsions during childhood. There was also no family history of epilepsy or febrile convulsions. Interictal electroencephalography (EEG) was performed about 7 hours after the generalized tonic–clonic seizure. He experienced nausea for about a minute during his EEG recording. He also manifested oral automatisms like swallowing and partial responsiveness during the attack. At that time, EEG showed an ictal period characterized by rhythmic sharp theta wave activity in the left temporal region lasting 80 seconds. Although his CT scan was normal, cranial MRI was performed a week later due to lateralization of EEG findings. On his cranial MRI scan, the left amygdala appeared edematous and left occipital medial cortical dysplasia was observed (Fig. 1).

He was advised to bathe with warm water for short periods and was prescribed 600 mg daily of oxcarbazepine. He has not had a recurrence during bathing or spontaneously for 12 months.

2.2. Patient 2

A 17-year-old boy presented with a 1-year history of seizures while bathing. The patient described how he would suddenly feel that the surrounding environment had become cloudy in the bathroom. This was followed by impairment of consciousness lasting about 1–2 minutes. No members of his family had witnessed his attacks. These attacks occurred three times a year while he was bathing. He hurt his foot and head during two episodes. He used to bathe by pouring hot water over his head from a bucket. When he bathed with warm water for a short time, he did not experience these attacks.

The patient had not experienced spontaneous seizures, but he had had severe headaches for a month and his right



Fig. 1. Cranial MRI findings for patient 1 with left occipital medial cortical dysplasia.

shoulder demonstrated slight weakness on neurological examination. There was no history of head injury or seizures during any febrile illness. However, his sister had experienced febrile convulsions during childhood.

Interictal EEG was normal but his cranial MRI showed a $6 \times 9.9 \times 8$ -cm arachnoid cyst in the left temporal region that caused shifting of the ventricles (Fig. 2). He was operated on within a month with shunting, and antiepileptic treatment was started with oxcarbazepine 600 mg daily. Unfortunately, we were able to follow him up for only a month. During that month after surgery, no seizures were observed.



Fig. 2. Cranial MRI findings for patient 2 with a huge arachnoid cyst in the left temporal region.

2.3. Patient 3

A 36-year-old man was admitted to our hospital for a new type of seizure, which he began to experience at the age of 34. The seizures were characterized by an aura with an abdominal feeling and *déjà vu*, followed by unresponsiveness for about 45 seconds. These attacks were provoked while bathing by pouring hot water over his head or when traveling by bus on a hot day. He reported feeling pleasure before some seizures, but he did not provoke the seizures. Usually a feeling of sadness followed the seizures.

The patient had had his first seizure spontaneously at 22 years of age; it was characterized by confusion after nausea. Neuroradiologic investigation had revealed a low-grade tumor in the right occipital lobe and he underwent surgery in another medical center. Unfortunately, further details on this attack and the operation were not available. After the operation, once or twice a month, he experienced spontaneous attacks like those experienced in the hot bathroom. For the last 2 years, almost 80% of his seizures had occurred during bathing.

When he attended the epilepsy clinic, at 36 years of age, he had been taking lamotrigine 200 mg daily and carbamazepine 800 mg daily. His neurological examination was normal. Cranial MRI showed sequelae in the right parieto-occipital area operated on and right hippocampal sclerosis. Because of deterioration in liver function and insufficient control of seizures, carbamazepine was stopped gradually and levetiracetam 1000 mg daily was added to the lamotrigine. He changed his bathing habits as well. He was seizure-free for 11 months during follow-up.

2.4. Patient 4

A 25-year-old woman was admitted to our hospital with an 8-year history of recurrent episodes of strange feelings while bathing. The patient described feeling as though she were in an alien place with a moving floor or as if she were late for something. Sometimes she remembered past bathing experiences during these seizures. Although unconscious, she had been walking around her home for few minutes. Pouring hot water over her head and face while showering triggered these attacks. She did not induce these seizures and derived no pleasure from them. Two years after the initial event, she had a similar spontaneous seizure.

Her past history revealed that she had had a febrile convulsion at the age of 1.5 years, but this was not certain. In addition, she had had enuresis nocturna until the age of 13. There was no family history of epilepsy or febrile convulsions. Neurological examination and interictal EEG were normal. However, cranial MRI showed right hippocampal atrophy (Fig. 3). Oxcarbazepine 900 mg daily was prescribed, and bathing with warm water was advised. She experienced no seizures during the 14-month follow up.

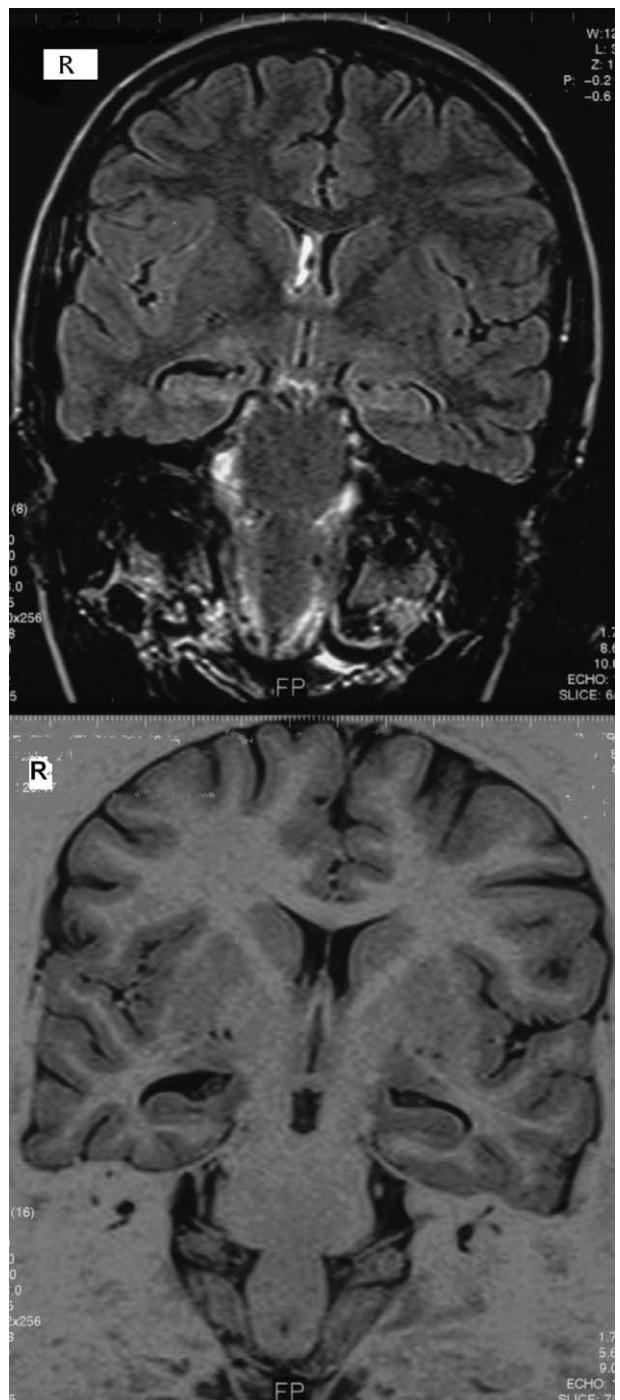


Fig. 3. Cranial MRI findings for patient 4 with right hippocampal atrophy.

2.5. Patient 5

A 16-year-old boy presented with a 3-year history of seizures while bathing. At the end of a long shower, he would have a generalized tonic-clonic seizure. The patient had had only six seizures while bathing and no spontaneous ones. His past history revealed a febrile seizure at the age of 2. Although there was no family history of seizures, his parents were consanguineous. He was mildly mentally

retarded. Interictal EEG showed diffuse slowing with 6–7 cycles/second. Cranial MRI demonstrated left hippocampal and parahippocampal atrophy.

Carbamazepine 600 mg daily was prescribed, and bathing with warm water was advised. He experienced no seizures during follow-up.

3. Discussion

According to the International League Against Epilepsy, HWE is included in the reflex epilepsies [16]. Although, in general, not common, HWE is concentrated in certain regions of the world [1,2]. The largest series of patients with HWE was reported by Satishchandra et al. [6]. After that, HWE was reported to account for 6.9% of all epilepsy cases in southern India, with a prevalence of 60/100,000 [1]. The second largest concentration of HWE cases is in Turkey, where the incidence was reported to be 0.6% by Bebek et al. [2]. There are also some isolated case reports from Australia, Canada, France, the United States, and Japan [5,7,9,12].

The reason why HWE occurs infrequently in Western cultures is not known. It may be due to genetic predisposition or bathing habits. Familial HWE cases with more than one affected member have been reported in 18% of the patients from India [1] and in 10% of Turkish patients [2]. The frequency of consanguineous marriage is high in Turkey (20%) [17] and India (18%) [18]. It has been postulated that autosomal recessive mutation is a cause of HWE. The coexistence of HWE and febrile seizures in the same family has also been reported [1,2], as in patients 2, 4, and 5 in this study. There may be no causal link between febrile seizures and HWE, but it may indicate a genetic predisposition. Traditional factors may also affect the occurrence of HWE; bathing habits such as using very hot water and pouring water from a bowl onto the head are common in these cultures [1,2].

The fact that complex partial seizures are the most common clinical presentation (67–80%) and ictal and interictal EEG recordings show an epileptic focus in the temporal lobe [1,3,7,10–12] suggested the presence of a structural lesion in the temporal lobe [10]. Although imaging data from CT or MRI were not available for all patients with HWE in these studies [2,3,6], there were no demonstrable structural changes on the MRI or CT scans, but functional involvement of medial temporal lobe structures and hypothalamus was suggested by one SPECT study [19].

The physiopathogenic mechanisms are usually complex in all types of reflex epilepsies. The cerebral cortex corresponding to the function that induces an epileptic crisis is hyperexcitable, and the cause may be an identifiable lesion or dysfunction without an underlying lesion [20]. To the best of our knowledge there are only three case reports of structural cerebral lesions associated with HWE in the literature. The first, reported from Japan, was an adult with HWE accompanied by pineal cyst and cavum septum pelucidi [13]. This was not explained as the cause of HWE

but may have occurred coincidentally with another structural lesion with HWE, as in our patient 2. Patient 2 had HWE and headaches. An arachnoid cyst in the left temporal lobe was demonstrated on MRI. Involvement of the temporal lobe might have been important in the development of HWE in that patient, because, following a shunt operation, he experienced no seizures when bathing with hot water and no headaches. The temporal lobe was also involved in the second case report of HWE with a cerebral lesion in the literature [12]. In this man, HWE was provoked by immersing his right hand into hot water. It was postulated that the coexistence of a cortical lesion over the left temporoparietal region adjacent to the sensory cortex initiated ictal EEG discharges. Involvement of the sensory cortex was also suggested in another case report of a 4-year-old girl in whom complex partial seizures were triggered by bathing in hot water. Interictal EEG showed epileptiform discharges in the left parietal area, and cranial MRI showed left parietal cortical dysplasia [14]. In our study, patient 1, with left occipital medial cortical dysplasia, is the third patient with HWE in whom a cortical malformation has been observed.

Involvement of the temporal lobe in HWE has also been postulated in animal models. Experimentally repeated exposure of the heads of adult rats to hot water increased susceptibility to convulsions. Ischemic changes in specific areas like Sommer's sector in the hippocampus, layer 4 and 5 neurons of the cerebral cortex, and reticular neurons in the brainstem were revealed in a pathological study. This was shown to have a kindling-like effect, mostly on the amygdala, and is called *hyperthermic kindling* [21,22]. It was also suggested that the seizures in HWE are a type of hyperthermic seizure similar to febrile seizures [22].

In addition to these experimental studies, Satishchandra et al. [23] postulated that patients with HWE probably have a genetically determined aberrant thermoregulatory system and are probably sensitive to rapid rises in temperature. They also reported that in these patients, it takes longer for body temperature to return to baseline after bathing compared with normal healthy volunteers. Although the role of the temporal lobe or amygdala was impressive in experimental animal models [21,22], there was no explanation of the involvement of these structures in the human study [23]. Our first patient had an edematous amygdala in addition to left occipital medial cortical dysplasia on cranial MRI performed a week after his seizure. Ictal EEG findings were also concordant with MRI findings. It was not clear whether these structural lesions were related to HWE or just coincidental findings, but the edematous appearance of the amygdala on MRI might be a subacute finding of seizure [24]. Most reports suggest focal cerebral edema as the basis of the transient imaging abnormalities. Therefore, the edematous appearance of the amygdala might be a sign of MRI changes after a seizure and thus indicate the epileptogenic area [25,26].

All these experimental studies with animals and studies with humans suggest that HWE probably has a genetic

basis and may have a kindling effect on hippocampus as do febrile seizures. Spontaneous seizures and a hippocampal pathology such as sclerosis are not rare in repeated febrile seizures. Due to the similarities between febrile seizures and HWE, it may be assumed that HWE may also cause spontaneous seizures and hippocampal sclerosis. Spontaneous seizures have been reported to occur a few years later in 16–38% of patients with HWE [1,2]. However, a case of spontaneous nonreflex seizures before HWE has also been reported [3]. It is not the case that all HWE patients with spontaneous seizures must have hippocampal pathology. Structural changes may develop as a result of the kindling effect of many HWE seizures and cause patients to have spontaneous seizures. The hippocampal sclerosis and spontaneous seizures in patient 4 may be explained by this hypothesis.

Spontaneous nonreflex seizures before and after HWE may also be a sign of a cortical hyperexcitable lesion like a tumor or dysplasia overlapping or coinciding with areas physiologically activated during sensory stimulation, as in patients 1 and 2. Patient 3's seizures may have a dual pathology. The first is a cortical hyperexcitable lesion like a tumor, overlapping or coinciding with areas physiologically activated during sensory stimulation. The second is a kindling effect on the hippocampus like hippocampal sclerosis. We were not able to test this idea, however, due to the lack of information about his surgery.

Patient 5, with hippocampal sclerosis without spontaneous seizures, could not be included in these groups. In this patient temporal injury may be associated with a febrile seizure history. This explanation may also be relevant to patient 4, although she had spontaneous nonreflex seizures.

In addition to the occurrence of nonreflex spontaneous seizures, the age at onset of HWE is important for the evaluation of patients and treatment, because self-limited HWE is seen mostly in the first decade of life in Turkey [2,3]. In the series described by Satishchandra et al., the mean age at onset of reflex seizures was 13.4 years [6]. Accordingly, late-onset HWE with spontaneous nonreflex seizures requires antiepileptic drugs in addition to a change in bathing habits.

HWE may represent the tip of an iceberg. Bathing with hot water may provoke seizures in some symptomatic focal epilepsies without the patients' realizing and reporting it. Furthermore, HWE is generally known to be benign and self-limited with precautions against precipitating factors, but it may be seen with spontaneous seizures. Antiepileptic drugs may sometimes be necessary to control seizures like those of our patients. In addition, there may be intracranial pathology like hippocampal sclerosis, dysplasia, and a huge cystic lesion requiring surgery. Therefore, rarely performed neuroimaging studies must be considered in the evaluation of these patients, especially those with late onset and spontaneous nonreflex seizures. Furthermore, investigations with new detailed neuroimaging techniques in addition to experimental and clinical studies might be helpful in understanding the mechanism of this reflex epilepsy.

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