



Hot Water Epilepsy in Children: A Rare Form of Reflex Epilepsy

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ABSTRACT

Aim: We evaluated the clinical and electroencephalography (EEG) characteristics, treatments, and outcomes of children with hot-water epilepsy (HWE), a specific type of reflex epilepsy.

Materials and Methods: This retrospective study included 11 children who were followed-up for HWE in a pediatric neurology department between 2005 and 2022.

Results: Eight children (73%) were boys and three (27%) were girls. The mean age was 60.7 ± 30.8 months (range 11-110) and the mean age at seizure onset was 57 ± 31.7 months (range 11-108). The mean follow-up duration was 20 months (range 10-32 months). The seizure type was identified as focal onset impaired awareness in six cases (54%) and generalized tonic-clonic seizures in five patients (46%). Four (36%) patients experienced spontaneous seizures. Interictal EEG was abnormal in two patients (18%). Four patients with spontaneous seizures were initially recommended bathing with lukewarm water and continuous anti-seizure medications. Three patients with reflex seizures benefited from lukewarm bathing and achieved seizure control. Two patients with uncontrolled reflex seizures were seizure-free after clobazam prophylaxis. Two patients who were unresponsive to lukewarm bathing and clobazam were started on continuous anti-seizure medications. In total, six patients were on continuous anti-seizure medications.

Conclusion: Despite its benign nature, HWE should be identified and appropriately treated due to the risk of spontaneous seizures. It is also important to determine the triggering factors so that appropriate bathing with lukewarm water and intermittent clobazam prophylaxis can be initiated. Spontaneous seizures require anti-seizure medications.

Keywords: Hot water epilepsy, children, clobazam

Introduction

Seizures can be precipitated by certain stimuli, including hot water and visual stimuli, in approximately 5% of epilepsy patients and these are defined as reflex epilepsies (1). Hot water epilepsy (HWE) is a specific type of reflex epilepsy which occurs with the pouring of hot water over the head. It was first described in 1945 in New Zealand (2). It is most common in South India, and is reported to

constitute 3.6-3.9% of epileptic seizures (3). Turkey is one of the countries where HWE is most frequently reported (4-7). Its etiopathogenesis is not clear, but genetic and environmental factors and bathing habits with hot water have been implicated (8).

In this study, we evaluated the clinical and electroencephalographic (EEG) characteristics, treatments, and outcomes of children with HWE.

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Materials and Methods

This study included 11 children who were followed up for HWE in the pediatric neurology department between 2005 and 2022. Data were retrospectively retrieved from the hospital and patient records, including patient characteristics, age at seizure onset, seizure semiology, the presence of spontaneous seizures, the presence of febrile seizures, family history of epilepsy, developmental status, duration of follow-up, response to modification of bathing habits, and antiepileptic treatment. Neurological and cardiac examinations, laboratory results, imaging, and EEG findings were evaluated. The history of seizures was based on the accounts of family members. This study was approved by the Institutional Review Board of Koç University (approval no.: 2023.077.IRB1.025, date: 09.03.2023). The analysis and reporting of the results complied with the strengthening the reporting of observational studies in epidemiology checklist.

Statistical Analysis

Data were analyzed using descriptive statistics and were expressed as number and percentages and mean±standard deviation, where appropriate.

Results

Eight of the children (73%) were boys and 3 (27%) were girls, with a male-to-female ratio of 2.6. The mean age was 60.7±30.8 months (range 11-110 months) and the mean age of seizure onset was 57±31.7 months (range 11-108 months). None of the patients had any history of febrile convulsions. Three patients (37%) had a family history of HWE, two of whom were siblings. Two (18%) patients had a family history of febrile convulsions. The mean follow-up duration was 20 months (range 10-32 months). Seven patients (64%) only had reflex seizures induced by hot water and four patients (36%) also had spontaneous seizures, which appeared after an interval of six to 12 months.

Seizure type was identified as seizures characterized by focal onset impaired awareness in six cases (54%) and generalized tonic-clonic seizures in five patients (46%), followed by post-ictal headache in one patient. All of the patients had normal development and neurological examination findings.

Brain magnetic resonance imaging (MRI) was unremarkable in all patients. Interictal EEG recordings were normal in nine patients (82%). The abnormal EEG findings in two patients (18%) included sharp wave activity in the temporal part of the right hemisphere in one patient

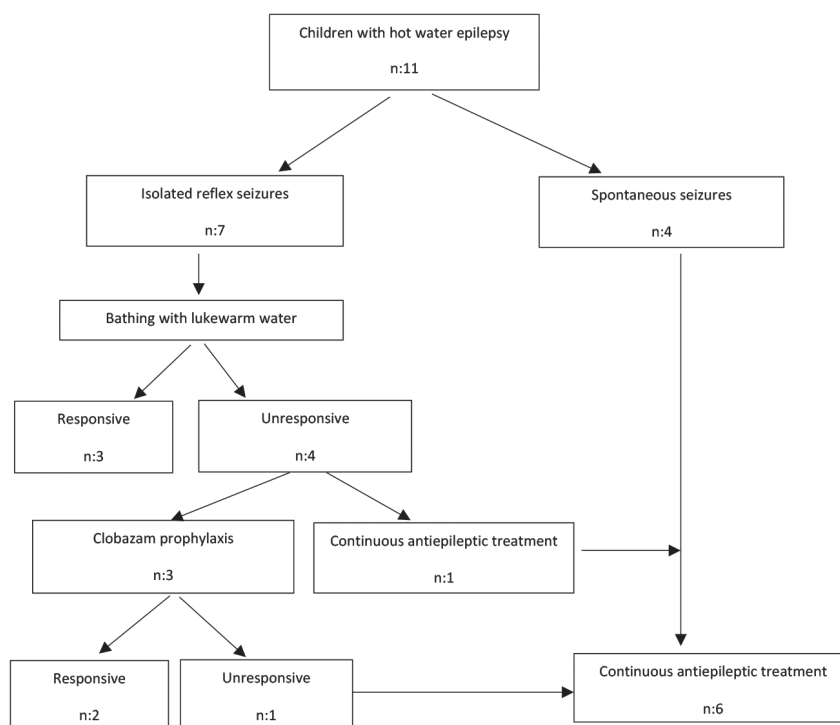


Figure 1. Flow chart of management of patients with hot water epilepsy

(Patient 11) and slow-wave discharges in the left temporo-occipital region in the other (Patient 7).

Treatments

Those patients with reflex seizures and spontaneous seizures were initially recommended to bathe with lukewarm water (Figure 1). Continuous antiepileptic treatment was initiated in those patients with spontaneous seizures. Of the seven patients with reflex seizures only, three benefited from lukewarm bathing and had seizure control. Three patients with uncontrolled reflex seizures received clobazam prophylaxis approximately one hour prior to bathing, of whom two had seizure control. Two patients who initially had reflex seizures but were unresponsive to

lukewarm bathing or clobazam were started on continuous antiepileptic treatment. In total, six patients were on continuous anti-seizure medications.

Anti-seizure medications included carbamazepine (n=4), levetiracetam (n=1), and valproic acid (n=1) (Table I).

One patient who received carbamazepine for spontaneous seizures also received clobazam prophylaxis due to the persistence of hot water-induced seizures. Five patients underwent continuous antiepileptic treatment. One patient (Patient 6) who discontinued carbamazepine after four months of antiepileptic treatment remained seizure-free during follow-up without any anti-seizure medications (Figure 1).

Table I. Patients' characteristics

Patient no	Gender	Age of onset (months)	Age at presentation (months)	Family history	Seizure semiology	Presence of spontaneous seizures	EEG	Follow-up (months)
1	F	24	28	HWE in a sibling	Generalized	+	Normal	24
2	M	108	108	No	Focal onset impaired awareness	+	Normal	16
3	M	36	48	No	Generalized	-	Normal	18
4	M	42	48	HWE in a sibling	Generalized	-	Normal	30
5	F	62	64	SSE	Focal onset impaired awareness	+	Normal	17
6	M	106	110	Febrile convulsion	Generalized	-	Normal	32
7	M	11	11	No	Focal onset impaired awareness	-	Abnormal	20
8	M	46	48	No	Focal onset impaired awareness	+	Normal	10
9	F	60	66	No	Focal onset impaired awareness	-	Normal	24
10	M	46	50	Febrile convulsion	Generalized	-	Normal	14
11	M	87	87	No	Focal onset impaired awareness	-	Abnormal	15

EEG: Electroencephalography, HWE: Hot-water epilepsy, CBZ: Carbamazepine, CLB: Clobazam

Table I. Continued

Patient no	Time interval between spontaneous and reflex seizures (months)	Response to lukewarm bathing	Antiepileptic medications	Prognosis
1	8	-	VP	A single spontaneous seizure during non-adherence.
2	10	-	CBZ, CLB	Seizure control following addition of intermittent CLB to CBZ for ongoing hot water seizure
3		+	-	Seizure-free
4		+	-	Seizure-free
5	12	-	CBZ	Seizure-free
6		-	CBZ	No seizures 4 months after discontinuation of CBZ
7		-	CLB, CBZ	No seizures after addition of CBZ to intermittent CLB
8	6	-	LVT	Seizure-free
9		+	-	Seizure-free
10		-	CLB	Seizure-free
11		-	CLB	Seizure-free

EEG: Electroencephalography, HWE: Hot-water epilepsy, CBZ: Carbamazepine, CLB: Clobazam

Discussion

HWE is a benign form of reflex epilepsy with a relatively favorable response to treatment and prognosis. However, antiepileptic treatment may be required in the presence of spontaneous seizures and/or unresponsiveness to non-pharmacological measures.

According to the largest study from India with 279 cases, HWE was more common in children and among males than among females (male to female ratio of 2.6/1) (9). In our study, there was a male preponderance. In the literature, the age of onset of HWE varies extensively from infancy to adulthood. Satishchandra (9) reported the most frequent age of onset to be 1-5 years. In our study, the age at onset ranged from 1 to 12 years.

HWE is often benign in nature and the modification of bathing habits may suffice. In more severe cases with or without spontaneous seizures, pharmacological treatment is required. Moreover, spontaneous seizures may develop if left untreated, which is reported in 17-25% of patients (10). Satishchandra (9) reported the development of spontaneous seizures in 25.4% of cases within 1-3 years. The prevalence of spontaneous seizures may be as high as 35.3% (11). In our study, spontaneous seizures accompanied HWE in 36% of the cases.

The presence of febrile seizures, as well as a family history of epilepsy and febrile seizures, has been reported in cases of

HWE (2,9,11). Satishchandra (9) reported febrile convulsions in 7% of their cases, a family history of epilepsy in 22.6%, and HWE in 7%. Meghana et al. (2) found febrile seizures in 14.2% of patients and a family history of epilepsy in 24.2% (HWE in 8%). In another study, 7.3% of patients had a family history of febrile seizures (11). In our study, three (37%) patients and two (18%) patients had a family history of HWE and febrile convulsions, respectively. Two patients with a family history of HWE were included in this study.

Concerning the semiology and EEG findings of HWE, Meghana et al. (2) reported focal-onset seizures with impaired awareness in all cases, with 58.5% showing focal-to-bilateral spreading. In another study, generalized tonic-clonic seizures were reported in 33% (8). We observed focal-onset seizures in 54% of patients, and focal-to-bilateral spreading was observed in 46% of our patients.

Radiological imaging of the brain is usually unremarkable for structural lesions (8). In a study of 38 cases, only three cases had incidental findings, such as a subarachnoid cyst, cavum septum pellucidum, or mega cisterna magna, in which the MRI of the brain was normal (11).

In the majority of cases with HWE, the interictal EEG is normal, with only 15-20% exhibiting diffuse abnormalities. In addition, spike wave discharges originating from the anterior temporal regions have been recorded in some

cases (3,8,9). EEG abnormalities were more frequently reported in those patients with accompanying spontaneous seizures than in those with HWE alone (11). In our study, EEG abnormalities were found in only two patients (18%) as focal changes in the temporal region, both of whom were free from spontaneous seizures.

Bathing with lukewarm water is the first-line treatment for HWE. Satishchandra (9) and Meghana et al. (2) reported that 38% and 13.3% of cases, respectively, remained seizure-free merely by the modification of their bathing habits. In the current study, 43% of our patients became seizure-free after the modification of their bathing habits. Generally, the frequency of HWE seizures is associated with the frequency of exposure to hot water over the head. However, in 5-10% of these patients, seizures may develop even during bathing, without hot water being poured over the head. Interestingly, self-induced seizures have been reported in approximately 10% of patients who experience intense pleasure from driving themselves until a loss of consciousness by compulsive exposure to hot water (8).

With the understanding that the pathogenesis of HWE is hyperthermic, similar to febrile seizures, intermittent prophylaxis has been proposed as an alternative treatment method along with bathing with lukewarm water (8). Satishchandra et al. (10) observed that prophylactic administration of clobazam 1.5 hours before bathing could be effective in HWE. In another report, 74.2% of patients remained seizure-free after intermittent clobazam treatment, and in 6.1% of the cases, seizures were reduced by more than 75% (12). Similarly, 75% of our patients benefited from clobazam with the resolution of seizures. Although intermittent clobazam prophylaxis before bathing may be effective in HWE, patients with spontaneous seizures require anti-seizure medications (12).

Study Limitations

The main limitation of the present study is its retrospective design. Seizure semiology was mainly based on patients' accounts. The small size of the patients from a single center may prevent generalizability of the results.

Conclusion

In conclusion, although HWE is a benign reflex epilepsy, HWE seizures should be identified and appropriately treated as there is always a risk of spontaneous seizures. At presentation, it is also important to determine the triggering factors and distinguish HWE from other epilepsies, so that the appropriate treatment of bathing with lukewarm water and intermittent clobazam prophylaxis can be initiated. Use

of anti-seizure medications until seizure control is required in cases of spontaneous seizures.

Ethics

Ethics Committee Approval: This study was approved by the Institutional Review Board of Koç University (approval no.: 2023.077.IRB1.025, date: 09.03.2023).

Informed Consent: The study was waived from informed consent from patients due to its retrospective design.

Footnotes

Authorship Contributions

Surgical and Medical Practices: A.A.A., S.U., Concept: A.A.A., S.U., Design: A.A.A., S.U., Data Collection or Processing: A.A.A., S.U., Analysis or Interpretation: A.A.A., S.U., Literature Search: A.A.A., S.U., Writing: A.A.A., S.U.

Conflict of Interest: No conflict of interest was declared by the authors.

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