Pediatric Neurology 99 (2019) 76–81

Contents lists available at ScienceDirect

Pediatric Neurology

journal homepage: www.elsevier.com/locate/pnu

Clinical Observation

Ictal Video-Electroencephalography Findings in Bathing Seizures: Two New Cases and Review of the Literature

Arantxa Mosquera-Gorostidi, MD [a, 1], Gurutzi Azcona-Ganuza, MD [b, 1], María Eugenia Yoldi-Petri, MD [a], Iñaki García de Gurtubay, MD, PhD [b], Sergio Aguilera-Albesa, MD, PhD [a, *]

[a] Paediatric Neurology Unit, Department of Paediatrics, NavarraBioMed Research Group, Navarra Health Service Hospital, Pamplona, Spain
[b] Department of Neurophysiology, NavarraBioMed Research Group, Navarra Health Service Hospital, Pamplona, Spain

ARTICLE INFO

Article history:
Received 15 August 2018
Accepted 21 April 2019
Available online 26 April 2019

Keywords:
Bathing seizures
Hot water epilepsy
Reflex seizures
Bath-induced seizures
Reflex epilepsy
Ictal EEG

ABSTRACT

Introduction: Reflex bathing seizures are described during the course of bathing in water near body temperature. These seizures differ from other epilepsies characterized by bathing-induced seizures such as hot water epilepsy, but there are few well-described patients and only some of these have been documented by ictal video-electroencephalography.
Methods: Our objective was to characterize the clinical presentation of bathing-induced seizures demonstrated by ictal video-electroencephalographic recordings with water temperature below 38°C. We described two previously unreported infants and reviewed additional cases in the literature that fulfilled those criteria.
Results: Eighteen infants were indentified. They were predominantly male (72%), and the mean age of seizure onset was 15 months (one to 36 months). The most frequent seizure triggers included pouring water over the face and immersion. Seizures were of focal onset with loss of awareness and prominent autonomic symptoms. Ictal video-electroencephalography revealed delta-theta high-amplitude focal waves involving temporal and adjacent regions, with a rapid spread to the ipsilateral hemisphere or generalization. Avoiding known triggers usually controlled the seizures, but carbamazepine, valproate, and levetiracetam were also helpful. Neuroimaging was normal in all cases, and neurodevelopment was unaffected.
Discussion: Bathing seizures predominate in boys with an early onset and a benign self-limited course. The use of ictal video-electroencephalographic recordings in these cases leads to diagnosis and reveals individual differences in triggers.

Introduction

Bathing seizures refer to a specific type of reflex seizures triggered by bathing a susceptible child in water near body temperature (36°C to 37°C). These seizures seem to be different from those in hot water epilepsy (HWE), a condition with a strong genetic predisposition described in southern India and Turkey where ritual bathing involves repeatedly pouring hot water (ranging from 38°C to 55°C) over the child's head. Some cases have been associated with focal brain lesions in temporal or parietal lobes in both children and adults.[1-3] The pathophysiology of HWE may be relate to a combination of repeated pouring of water (kindling effect) and the high temperature of water during bathing in genetically or anatomically susceptible subjects.[2-4] In many cases of HWE, lowering water temperature is enough to avoid seizures. However, in bathing seizures water is usually at core temperatures and other triggers may be involved. A systematic review of the literature shows a confusing mixture of both types of seizures in published series. The number of reported infants with bathing seizures is small, and diverse methodologies for outlining the triggers and the semiology of seizures have been described.[1,5-15]

Financial disclosures: All authors stated that there were no specific financial interests, relationship, and affiliations relevant to the subject of the manuscript.
Conflict of interest: None.
* Communications should be addressed to: Aguilera-Albesa; Paediatric Neurology Unit; Department of Paediatrics; Navarra Health Service Hospital; c/ Irunlarrea, 4 Pamplona, 31080, Spain.
E-mail address: saguilea@navarra.es (S. Aguilera-Albesa).
[1] Both authors contributed equally to this work.

https://doi.org/10.1016/j.pediatrneurol.2019.04.017
0887-8994/

Ictal video-electroencephalographic (v-EEG) recording during specific conditions is useful for the delineation of reflex seizures. Although v-EEG recorded while pouring water over different parts of the body during a bath is not routinely used, it adds valuable information for the recognition of triggers and clinical features that may be difficult to achieve in other settings. Moreover, recordings also allow differentiating seizures from other bathing-induced paroxysmal disorders described in infants, such as alternating hemiplegia of childhood, hyperekplexia, and paroxysmal extreme pain disorder, in which episodes can be triggered with warm or cool water temperatures (30°C) in neonates.[1] Our objective was to describe the clinical and ictal EEG characteristics of seizures induced by bathing with water temperature below 38°C. Our hypothesis is that this subgroup represents a specific type of reflex seizures in respect to triggers, as it has been previously suggested by other authors.[1,10,14]

Methods

We described two new infants and review the other unambiguous cases from the literature. Articles were reviewed after a PubMed search using keywords bathing epilepsy, hot water epilepsy, reflex seizures, bath-induced seizures, reflex epilepsy, and ictal EEG. We included only cases with reflex seizures during a bath with water at temperature below 38°C and a detailed description of ictal v-EEG recordings available. We also considered only patients with a normal cerebral magnetic resonance imaging (MRI) study to differentiate genetically from anatomically susceptible subjects.[5-15]

Results

Patient 1: This 30-month-old boy was born to healthy nonconsanguineous parents after an uncomplicated delivery. His family history was unrevealing. Psychomotor development was normal. At age 10 months, he developed recurrent episodes of pallor, cyanosis, limpness, and loss of awareness that lasted one to two minutes, each time he was bathed. Since then, he refused to be bathed in the bathtub. Breath-holding spells were initially suspected, with a normal complete blood count, ferritin levels, and electrocardiogram. Baseline and sleep EEG including photostimulation were also normal. As all episodes were bath related, a v-EEG was performed while bathing the child. Sudden tactile, visual, and auditive stimuli did not produce any change in behavior or EEG pattern. Warm water at 36.5°C to 37°C was then poured over the legs, abdomen, and chest, shoulders, arms, neck, and back. EEG recordings began showing bitemporal 3- to 4-Hz waves at 46 seconds with no clear clinical changes for the next 49 seconds. The child suddenly developed pallor, cyanosis, diminished muscle tone, and a staring gaze; EEG showed generalized high-amplitude (300 μV) slow waves at 2 Hz. The EEG pattern normalized abruptly after 78 seconds, together with abrupt clinical recovery and crying (Fig 1). Cerebral MRI was normal. Bathing epilepsy was diagnosed, and valproate was initiated. Subsequently, the child remained episode free during bathing for the next 16 months. Unfortunately, seizures reappeared upon medication withdrawal prompting reintroduction of valproate.

Patient 2: This 13-month-old healthy girl, born to unrelated parents, developed episodes of cyanosis, limpness, and loss of awareness that lasted a few seconds and occurred only during routine bathing. Initial investigations were normal, including interictal EEG with photostimulation. v-EEG was performed during a bath. Sudden tactile, visual, or auditory stimuli did not produce changes in behavior or the EEG pattern. Likewise, pouring cool water over different parts of the child's body had no effect. However, 70 seconds after pouring water at 36.5°C over her face, an episode characterized by perioral cyanosis, head drop, and staring gaze took place. The episode lasted seven seconds and was followed by abrupt crying and spontaneous recovery. Ictal EEG recordings showed bilateral centrotemporal high-amplitude (150 μV) 4-Hz waves that normalized abruptly after seven seconds (see Fig 2). Neuroimaging was normal. Thereafter, bathing with warm wet clothes over the head and body was enough to avoid seizures.

Review of the literature: Only 16 cases presented bath-induced seizures with water at temperature below 38°C that were recorded by v-EEG. The clinical presentation and characteristics of the 16 cases described in the literature are detailed in Table.[5-15]

Discussion

Bathing seizures have not been fully characterized in the literature because ictal recordings are not systematically performed to support the diagnosis. This may occur because v-EEG recordings during a bath are difficult to obtain. Technical conditions are challenging because of the presence of water, especially in younger children. To better characterize these reflex seizures and to avoid attributing the episodes to nonepileptic paroxysmal events, we have described two new cases and identified 16 additional cases in

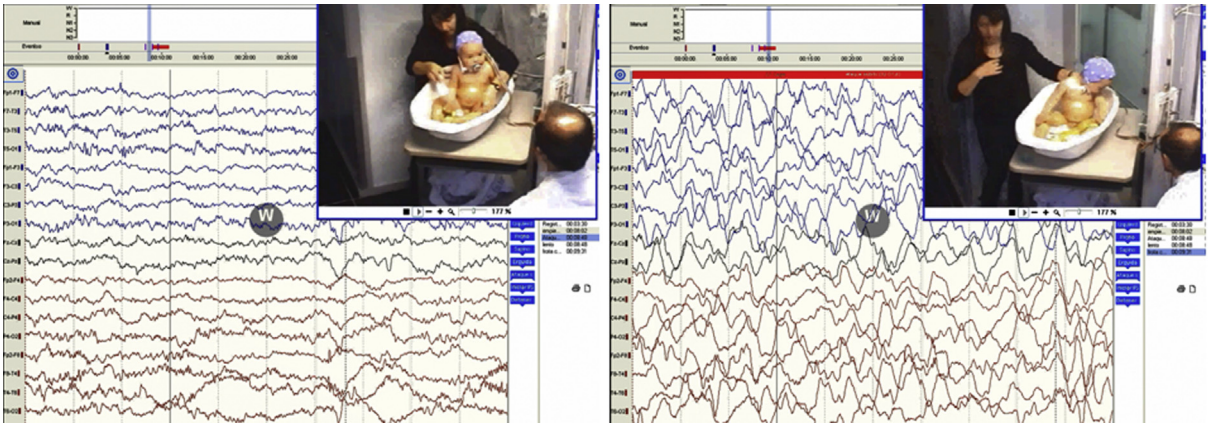

FIGURE 1. Patient 1. Electroencephalographic activity, 10-second epoch, 100 uv/cm, 30-month-old boy. Left: Basal EEG activity 6 to 7 Hz, 50 μV. Right: Two minutes after starting bath. Generalized delta 2 Hz, high-amplitude 300- to 400-μV waves. Clinically pallor, blue lips, limpness, and staring gaze. EEG, electroencephalography. The color version of this figure is available in the online edition. The video associated with this figure can be viewed at https://doi.org/10.1016/j.pediatrneurol.2019.04.017.

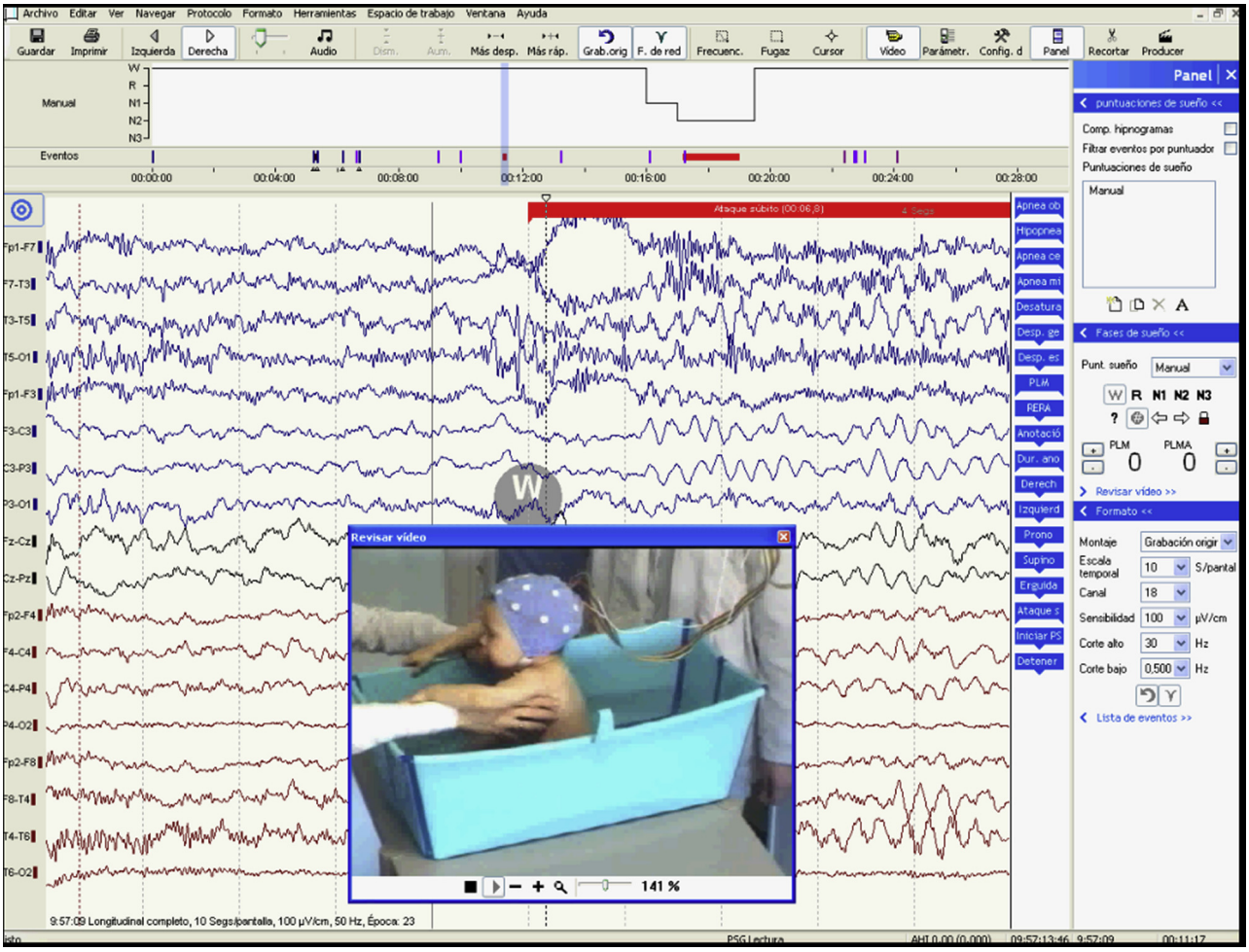

FIGURE 2. Video capture from Patient 2. Electroencephalographic and clinical description of the event. Time line in minutes and seconds. During the previous 10 minutes, water had slowly been poured consecutively over legs, abdomen, thorax, and shoulder, waiting for clinical changes at least for 90 seconds in the poured area. 0:00: Normal EEG with open eyes. Seventy seconds earlier we poured water over her face. 0:09: Bilateral centrotemporal high-amplitude (140 μV) 4-Hz waves appeared abruptly with head drop, staring gaze, and slight perioral cyanosis, for the next seven seconds. 0:16: Normalization of the EEG with abrupt crying and instantaneous clinical recovery. 0:22: A repetition of the video sequence is provided with increased video size to better characterize the clinical event. 0:26: Normal EEG. 0:32: EEG and clinical changes. 0:39: EEG and clinical recovery. EEG, electroencephalography. The color version of this figure is available in the online edition. The video associated with this Figure can be viewed at https://doi.org/10.1016/j.pediatrneurol.2019.04.017.

the literature with bathing-induced seizures (less than 38°C) demonstrated by ictal v-EEG (Table).[5-15]

The combined evidence reveals a male predominance (72%). The mean age of symptom onset was 15 months (range zero to 36 months). Patients had no family history of epilepsy. Pouring water over the head and the face as well as immersion were the most frequently identified triggers. Shoulders, chest, or genitalia were also described as trigger areas. We recommend pouring water gently over the child, beginning at the lower extremities leading progressively to the face during the v-EEG, for a better identification of the somatosensory trigger area. Seizure latency, when documented, ranges from a few seconds to a minute. Seizure duration was short-lived averaging one to three minutes (range; seven seconds to 10 minutes).

In all 18 patients, the clinical and electric v-EEG description suggests that seizures were of focal onset, with impaired awareness and autonomic symptoms. At the beginning of the seizure, affected infants generally became quiet. Their silence was accompanied by cyanosis or blue lips, staring eyes, limpness, and loss of alertness. Stiffness, clonic movements of the lips or one part of the body, and oral automatisms were also observed in some children. Autonomic symptoms including pallor, vomiting, tachycardia, and apnea have also been reported. In one case, laughter was the initial manifestation of seizure activity. In our second case, head drop was observed at the beginning of the seizure, which we believe was probably related to partial loss of muscle tone. Postictal drowsiness or crying, with coughing and facial flushing, have been also informed.

Interictal EEG was usually normal, with occasional focal slow waves during sleep recordings. In most of the reviewed cases, ictal v-EEG recordings showed delta-theta high-amplitude focal waves involving temporal and adjacent regions, and also starting from parieto-occipital regions, followed by a rapid spread to the ipsilateral hemisphere or generalization.

Only three infants exhibited sporadic nonprovoked seizures (17%), and in another three, bathing-induced seizures were reported occasionally. The prognosis is thought to be relatively benign. Trigger avoidance usually controls seizures, but carbamazepine, valproate, and levetiracetam have been useful. Neurodevelopment is invariably normal.

Our review suggests that bathing seizures are a type of reflex seizures with an early onset and a self-limited course in most cases.

TABLE.
Patients Reported With Bathing-Induced Seizures With Water at Normal Body Temperature and Ictal Video-EEG Recordings Available

Reference	Age at Onset (Months)/ Gender	Water T[a]	Trigger	Semiology	Latency After Start Bathing	Duration of Seizure	Recovery	Interictal EEG	Ictal EEG	Treatment Effective	Outcome
Stensman et al.[5]	7/M	37°C-37.5°C	T[a]37°C was decisive. Neither sitting in hot water nor heating fan	Blue, stiff, eyes staring, automatisms, right arm, stiffness, coughing 1 min later and then red face	30 s	1-2 min	Tired, fall asleep	Normal or slow activity right postcentral	Initially, focal delta high amplitude left frontotemporal, followed by generalized slow waves (1-3 Hz)	CBZ	Occasional seizures
Shaw et al.[6]	5/M	Warm 37°C	Following immersion	Staring, cyanosis, pallor, and hypotonia	30 s	10 min	Sleep	Normal	High-amplitude delta activity over the left hemisphere	VPA	Free
Ioos et al.[7] case1	6/M	Warm 34°C-37°C	Following immersion	Unresponsive and hypertonic, eyes gazed upward, clonic movements of the lips	30 s	1-2 min	Cry and after that sleep	Normal	Theta-delta slow waves over right parieto-occipital and temporal regions with spreading to the left side	Water < 34°C	One additional seizure at 14 mo
Ioos et al. case 2	7/F	ND	ND	Floppy with her eyes gazing upward and cyanotic lips	ND	ND	ND	Normal	Rhythmic delta slow waves over the left hemisphere, which then spread to the right side	CBZ and bath stop	Nonprovoked seizures
Ioos et al. case 3	6.5/M	36°C shower	ND	Complex partial seizures	ND	ND	ND	Normal	Right temporal rhythmic slow waves, then generalized	Shower at 36°C and VPA	21 mo no seizures
Ioos et al. case 4	24/M	<38°C	ND	Complex partial seizures	ND	ND	ND	Normal	Right temporal rhythmic slow waves, then generalized	Water < 38°C and CBZ	CBZ needed
Ioos et al. case 5	7.5/M	<35°C	ND	Complex partial seizures	ND	ND	ND	Normal	Bilateral temporal slow waves, then generalized	<35°C	Free
Argumosa et al.[8]	Newborn/ F	Hot or warm	Pouring water over head	Cyanosis lips, clonic movements of the lips, loss of awareness, and hypotonia	ND	1-2 min	Abnormal steps and somnolence	Normal	Bilateral frontotemporal spikes	VPA	Free
De Keyzer et al.[9]	10/M	Warm (37°C)	After immersion	Glazed eyes staring, cyanosis, pallor, unresponsiveness, hypotonia	1 min	1 min	Drowsiness	Normal	Left temporal rhythmic slow wave activity of high amplitude rapidly spreading to the left frontal and right frontotemporal regions	ND	ND
Auvin et al.[10]	24/F	From 33°C	Only over head and face	Fear, left hypotonia, loss of consciousness	ND	16 sec	ND	Normal	Right frontoparietal high-voltage slow waves	OXC	Seizures > 6 y old
Ceulemans et al.[11]	13/M	Warm 36.6°C	While taking bath	Stopped playing, staring, pallor, perioral cyanosis, hypotonic	20 s	1 min and 30 sec	Fell asleep	Normal	Occipital slow waves with high voltage delta waves, and generalization	Lukewarm (32°C) and VPA	Free
Franzoni et al.[12]	4/F	Lukewarm	Pouring water over genitalia	Loss of consciousness, asymmetric right tonic phases, eyes to right, wandering gaze	5-30 s	2-4 min	Cry, prolonged drowsing	ND	Generalized high-amplitude waves > left central	Wet clothes bath	Free at age 7 y
Jansen et al.[13]	24/M	37°C	Shower over the head	Quiet, pale, nauseated, vomiting, oral automatisms, limpness, absent	Seconds	3 min	Sleep	Normal	Irregular delta left anterior temporal, then high amplitude spreading all temporal unilateral	CBZ: side effects LEV	Free with LEV
Kravljanac et al.[14] case 1	36/M	Lukewarm	Over body, not head	Motor arrest, hypotonia, staring, head drop, oral automatisms, perioral cyanosis	ND	2 min	Sleep 30 min	Only sleep frontal left	Generalized delta waves start left frontocentral, slow and high-amplitude waves	PB	One unprovoked seizure at age 3.8 y
Kravljanac et al. case 2	9/M	Lukewarm	Put face in water	Staring, confusion, right head deviation, immobility, oral-hand automatisms	ND	>68 s	somnolence	Sleep frontal right	Slow back, then 2-3 Hz start right temporal, generalized in 42 s, 68 s diazepam.	Bathing changes	Normal at age 3.5 y

(*continued on next page*)

TABLE. (*continued*)

Reference	Age at Onset (Months)/ Gender	Water T[a]	Trigger	Semiology	Latency After Start Bathing	Duration of Seizure	Recovery	Interictal EEG	Ictal EEG	Treatment Effective	Outcome
Stutchfield et al.[15]	19/M	Lukewarm	Over chest	Blue lips, hypotonia, tachycardia, brief apnea, stared straight ahead, mouthing movements	Seconds after pouring water over chest	2-3 min	Sleepy 1 h	Normal	Left temporal delta activity, spread right side	CBZ	Occasional unprovoked seizures Free with CBZ
Patient 1	30/M	36.5°C-37°C	Over legs, ascendant to shoulders	Pallor, cyanosis, limpness, staring gaze	95 s after pouring water over legs	29 s	Abrupt recovery, crying	Normal	Bitemporal 3- to 4-Hz waves, secondary generalization with slow 2-Hz waves	VPA	Relapse reflex seizures after VPA withdrawal
Patient 2	13/F	36.5°C	Over face	Perioral cyanosis, head drop, staring gaze	70 s	7 s	Abrupt recovery, crying	Normal	Bilateral centrotemporal high-amplitude 4-Hz waves	Bathing changes	Free at age 3 y

Abbreviations:
CBZ = Carbamazepine
F = Female
M = Male
ND = Not detailed
PB = Phenobarbital
VPA = Valproic acid

They present with a focal onset and predominantly nonmotor features including impaired awareness and autonomic symptoms, triggered by warm water at or near body temperature. These characteristics are slightly different from the originally reported HWE in patients from southern India or Turkey.[2,3] Recently, Meghana et al. reported 70 patients from a specific region in India with a mean age at onset of 18 years who presented with hot water epilepsy.[2] Seizures were triggered by hot water at 40°C to 55°C. Only 14% of the children had documented seizures when exposed to warm water under 40°C. Moreover, 36% of these children have spontaneous seizures, chief among them generalized tonic-clonic seizures.

Interestingly, a male predominance has been reported in both water-induced seizures, but with a diverse family history. In bathing seizures affecting infants, a male to female ratio of 2.5:1, with no preexisting family history appears to be the norm. In larger HWE series from India, the ratio is similar, but a family history of epilepsy (7% to 18%) is common.[2] Nguyen et al. described in 2015 seven of 10 patients belonging to a single family with reflex bathing-induced seizures, all of them males with a childhood onset (age four to 13 years), suggesting an X-linked pattern of inheritance. A mutation in the synapsin 1 gene on chromosome Xp11-q21 was demonstrated, but spontaneous seizures and language and social interactions deficits were also present in these individuals.[16]

Regarding the pathophysiology of water-induced reflex seizures, there is some evidence of temporoinsular and parietal network hyperexcitability.[16-18] A recent study using EEG-functional MRI in an adult with idiopathic HWE showed ictal abnormalities in left frontoparietal regions and also right superior temporal and parahippocampal gyrus.[17] In adults with secondary focal epilepsy, hot-water- and water-contact-induced seizures are related to structure abnormalities involving temporal and parietal lobes.[18] These authors suggest that there are regions of cortical hyperexcitability that overlap with areas physiologically activated during specific sensory stimulations. In addition, the SYN1 knockout mouse model showed that SYN1 mutations affect trafficking and produce an imbalance in release of synaptic vesicles, leading to a higher network excitability and firing activity.[16]

The accumulated evidence from different studies of water-induced seizures suggest that the experience of bathing may activate a genetically based hyperexcitability temporoinsular network that, when sufficiently activated, results in a focal seizure. This concept relies upon a series of clinically heterogeneous entities associated with water reflex seizures: idiopathic HWE, SYN1-related X-linked focal epilepsy, and secondary focal epilepsy in adults.[2,3,10,16-19] We suggest that the 18 cases summarized in Table may represent a specific type of reflex seizures as part of a male predominance spectrum of water-induced seizures, with an earlier age of onset and more benign prognosis. This profile may also suggest an age-related aberrant, genetically determined thermoregulatory system, as it is hypothesized in patients with HWE with an older age of onset, and also in patients with febrile seizures.[20]

In conclusion, our experience shows us that when an infant has repeated paroxysmal events during bathing, performing an ictal v-EEG recording during a normal bath should be considered. This recording will facilitate the identification of triggers and the characterization of the semiology of the episode. A correct diagnosis and the identification of triggers will help the family avoid seizures.

References

1. Nechay A, Stephenson JBP. Bath-induced paroxysmal disorders in infancy. Eur J Paediatr Neurol. 2009;13:203–208.
2. Meghana A, Sinha A, Sathyaprabha TN, Subbakrishna DK, Satishchandra P. Hot-water epilepsy clinical profile and treatment-a prospective study. Epilepsy Res. 2012;102:160–166.
3. Bebek N, Gürses C, Gokyigit A, Baykan B, Ozkara C, Dervent A. Hot water epilepsy: clinical and electrophysiologic findings based on 21 cases. Epilepsia. 2001;42:1180–1184.
4. Klauenberg BJ, Sparber SB. A kindling-like effect induced by repeated exposure to heated waters in rats. Epilepsia. 1984;25:292–301.
5. Stensman R, Ursing B. Epilepsy precipitated by hot water immersion. Neurology. 1971;21:559–562.
6. Shaw NJ, Livingston JH, Minns RA, Clarke M. Epilepsy precipitated by bathing. Dev Med Child Neurol. 1988;30:108–114.
7. Ioos C, Fohlen M, Villeneuve N, et al. Hot water epilepsy: a benign and unrecognized form. J Child Neurol. 2000;15:125–128.
8. Argumosa A, Herranz JL, Barrasa J, Arteaga R. Reflex epilepsy from hot water: a new case and review of the literature (Spanish). Rev Neurol. 2002;35:349–353.
9. De Keyzer K, Corthouts I, Van Coster R, Verhelst H. Hot-water epilepsy: a new Caucasian case. Eur J Pediatr. 2005;164:184–185.
10. Auvin S, Lamblin MD, Pandit F, Bastos M, Derambure P, Vallée L. Hot water epilepsy occurring at temperature below the core temperature. Brain Dev. 2006;28:265–268.

11. Ceulemans B, Koninckx M, Garmyn K, Wojciechowski M. Hot water epilepsy: a video case report. Epileptic Disord. 2008;10:45–48.
12. Franzoni E, Gentile V, Grosso S, Brunetto D, Cordelli LM, Balestri P. Bathing epilepsy: report of two Caucasian cases. Epileptic Disord. 2010;12:88–90.
13. Jansen K, Vervisch J, Lagae L. Bathing epilepsy: a video case of an autonomic seizure. Epileptic Disord. 2010;12:236–238.
14. Kravljanac R, Djuric M, Milovanovic M, Radivojevic V. Epileptic seizures provoked by bathing with water at room temperature. Epileptic Disord. 2012;14:321–324.
15. Stutchfield CJ, Loh NR. Focal epilepsy presenting as a bath-induced paroxysmal event/breath-holding attack. Epilepsy Behav Case Rep. 2014;2:102–104.
16. Nguyen DK, Rouleau I, Sénéchal G, et al. X-linked focal epilepsy with reflex bathing seizures: characterization of a distinct epileptic syndrome. Epilepsia. 2015;56:1098–1108.
17. Sandhya M, Bharath RD, Panda R, et al. Understanding the pathophysiology of reflex epilepsy using simultaneous EEG-fMRI. Epileptic Disord. 2014;16:19–29.
18. Sala-Padró J, Toledo M, Sarria S, et al. Reflex seizures triggered by cutaneous stimuli. Seizure. 2015;33:72–75.
19. Peron A, Baratang NV, Canevini MP, Campeau PM, Vignoli A. Hot water epilepsy and SYN1 variants. Epilepsia. 2018;59:2162–2165.
20. Italiano D, Ferlazzo E, Gasparini S, et al. Generalized versus partial reflex seizure: a review. Seizure. 2014;23:512–520.