■ Clinical commentary

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Bathing epilepsy: a video case report

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ABSTRACT – Bathing epilepsy is a rare form of reflex epilepsy triggered by bathing in room temperature water. It predominates in boys with a mean age of 15 months and its evolution is benign. Diagnosis of bathing epilepsy requires the exclusion of other paroxysmal disorders triggered by water contact. Video-EEG confirmation of the seizures is necessary to reach a diagnosis of certainty and to allow adequate management. We present the case of a one-year-old boy who experienced recurrent episodes of unresponsiveness and cyanosis while bathing in lukewarm water. The diagnosis of bathing epilepsy was confirmed by the video-EEG recording of a seizure, showing left-sided frontotemporal delta activity with rapid contralateral spread. Therapy with levetiracetam was effective, subsequently allowing bathing without further seizures.

Key words: reflex seizures; bathing; infant; video; EEG

Bathing epilepsy is a rare entity and its differential diagnosis is extremely broad [1]. A diagnosis of certainty is reached only through careful history taking and by means of a video-EEG recording of the paroxysmal event [2]. Although the prognosis of bathing epilepsy is favorable, a clear identification of this condition is necessary to implement alternative bathing techniques and to set adequate therapy in selected cases [3].

Case report

A one-year-old boy was admitted to our hospital due to recurrent episodes of cyanosis and unresponsiveness that began soon after immersion in the bath and lasted for a few minutes, followed by sleepiness for about one hour. The episodes started at the age of 10 months and occurred every time the child came in contact with water, with no specific association with exposed body area or water temperature. Two similar events occurred on breastfeeding and one in the absence of triggers. Between the paroxysmal events, the child had been well.

Medical history was unremarkable, and family history was negative for epilepsy and febrile seizures. Clinical examination was normal, as well as standard EEG in awake and sleep states, and brain MRI. Routine blood tests were unremarkable. An attempt was undertaken to provoke an episode by placing the child in water (36°C) whilst performing EEG and ECG with video monitoring. The EEG preceding the seizure showed normal background activity (*figure 1A*). Twenty seconds after seated immersion in water, a build-up of left-sided frontotemporal delta activity interspersed with sharp-waves appeared on the EEG recording (*figure 1B*), which soon involved the right side (*figure 1C*). Shortly after the EEG onset over the left frontotemporal areas, the child showed repetitive movements of the left upper limb, that could represent ictal motor automatisms ipsilateral to the EEG focus. The right arm appeared anchored in a fixed position

VIDEO ONLINE

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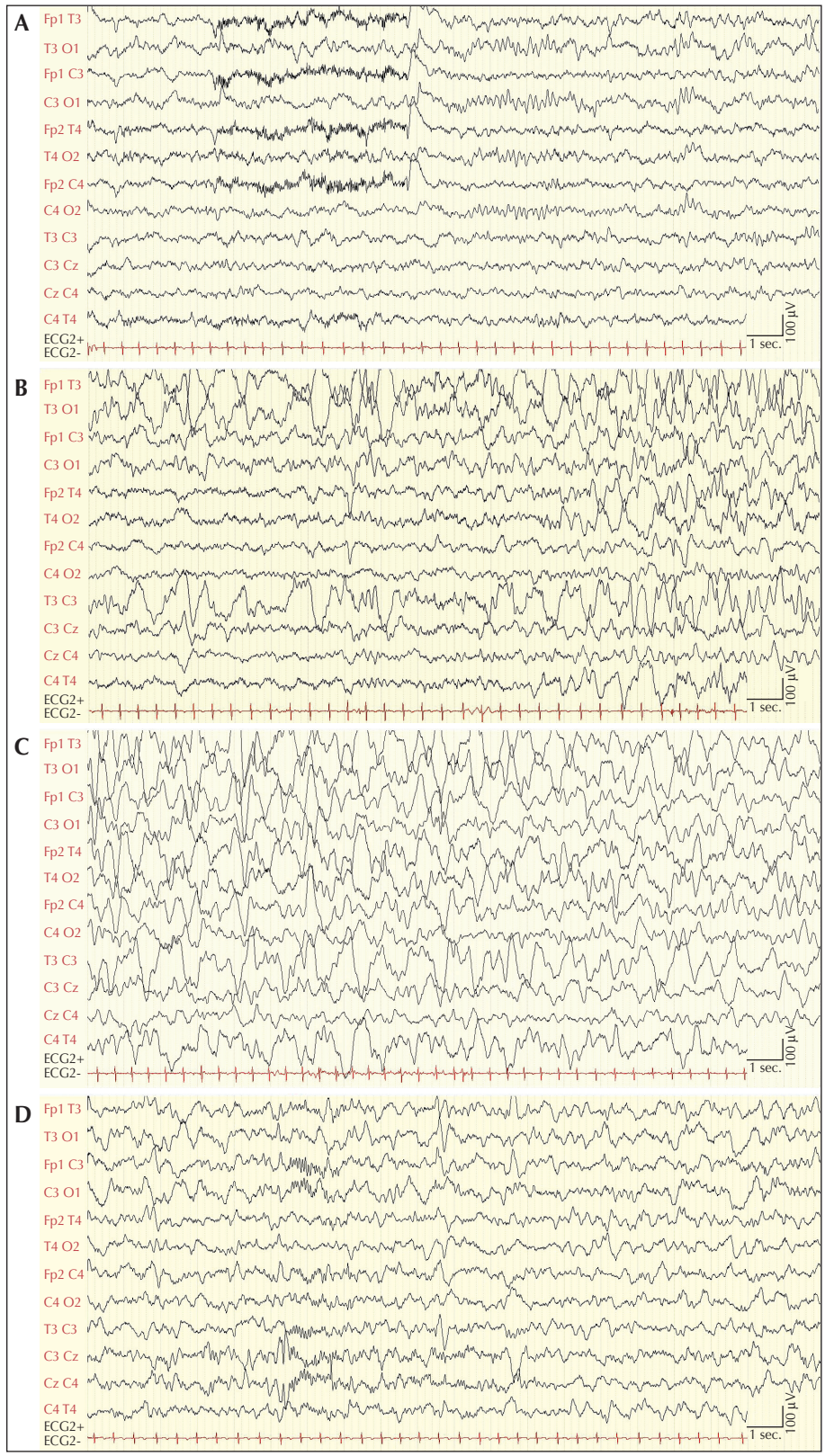

■ **Figure 1.** (A) Before seizure onset: normal awake tracing. (B) Seizure onset: build-up of left-sided frontotemporal delta activity interspersed with sharp waves. (C) Seizure spreading to the right side. (D) After seizure: normal sleep tracing.

to the edge of the bathtub, suggesting a contralateral dystonic posture. After 25 seconds, the child became motionless, unresponsive, with closed eyes and perioral cyanosis. Oxygen saturation dropped to 57% for a few seconds, followed by progressive spontaneous return to normal values. Continuous ECG monitoring showed a stable sinus rhythm throughout the whole recording, and no sinus bradycardia was encountered during the desaturation event. One minute later, the child regained alertness, although he moved poorly, and fully recovered with crying behavior within a few minutes. Soon after, the child fell asleep and the EEG showed a normal sleep pattern (*figure 1D*).

Treatment with levetiracetam was started at 10 mg/Kg/d and bathing avoidance was suggested in the home setting. In the following weeks, levetiracetam was progressively increased to 50 mg/Kg/d and bathing was possible without further seizures. After four weeks, a provocation test was carried out again, and neither paroxysmal events nor EEG discharges were detected.

Discussion

Water-induced seizures fall as a group under the category of reflex seizures, which are defined as epileptic events triggered by specific stimulation, namely a visual, auditory, cognitive or somatosensory stimulus [4].

The first step in the diagnostic approach is to confirm the epileptic nature of the event, excluding some non-epileptic conditions such as syncope, breath-holding spells and less common disorders, such as alternating hemiplegia of childhood (AHC), hyperekplexia and paroxysmal extreme pain disorder (PEPD). In our case, syncope and breath holding spells were ruled out because of the prolonged post-ictal state, absence of distress from the child and lack of ECG abnormalities [2]. Furthermore, no abnormal ocular movements, dystonia or hemiplegia were reported during the episodes, making the hypothesis of AHC unlikely [5], and no startle reaction or hypertonic stiffness were appreciated, ruling out hyperekplexia [6]. Finally, the absence of signs of pain perception by the infant, such as skin flushing, stiffening or screaming, excluded PEPD [7].

The second step is to discriminate between different forms of epilepsy associated with water contact. Water temperature plays a crucial role in this purpose, allowing a distinction between hot water epilepsy (HWE) and bathing epilepsy. HWE is a specific clinical entity, mostly described in southern India and Turkey where ritual bathing is common and involves the repeated pouring of hot water (40°-50°C) over the head [8,9]. Water temperature and the direct water pouring over the head are believed to be the main triggering factors, since paroxysmal events cannot be evoked if the water temperature is lowered [8,10]. In our case, the temperature of the water was lukewarm and there was no clear relationship with the pouring of water over the head, thus excluding HWE.

On the other hand, bathing epilepsy is a rare benign entity that predominates in boys with a mean age of 15 months. Bathing seizures have been reported in children up to three years old [3]. Patients have a normal clinical examination and psychomotor development [11]. Only 18 infants with this condition confirmed by ictal video-EEG recordings have been reported in the literature. Seizures are mainly characterized by quietness, cyanosis, staring, limpness, and loss of alertness. Stiffness, clonic movements, oral automatisms and autonomic symptoms have also been reported. In three out of the 18 reported cases (17%), sporadic non-provoked seizures also occurred. Ictal video-EEG often reveals delta-theta high-amplitude focal waves involving temporal and adjacent regions, rapidly spreading to the ipsilateral hemisphere or generalizing. Interictal EEG is usually normal, with occasional focal slow waves during sleep recordings [3]. In our case, clinical features and EEG ictal findings were consistent with bathing epilepsy.

The differential diagnosis of bathing epilepsy is complex and a comprehensive workup, including a video-EEG recording of the seizures during bathing, is necessary to exclude non-epileptic events or other epilepsy syndromes [2,3].

The prognosis of bathing epilepsy is usually benign. Data on treatment and outcome are available for 17 of the 18 reported cases. Changes in bathing only, such as lowering water temperature or using wet cloths, were adopted in five patients; four were free of seizures at follow-up, while one additional seizure was reported in one case. In the remaining 13 cases, antiseizure medications (ASMs) were started. Carbamazepine was the most used medication (five cases), followed by valproate, levetiracetam, phenobarbital or oxcarbazepine. No further seizures were reported in seven cases, while sporadic reflex seizures were described in the others, either during therapy or after ASM withdrawal. In two cases receiving pharmacological treatment, occasional non-provoked seizures occurred. Neurodevelopment was normal in all the reported cases [3]. ■

Supplementary material.
Summary slides accompanying the manuscript are available at www.epilepticdisorders.com.

Disclosures.
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Legend for video sequence

The video sequence shows an electroclinical seizure characterized by left upper limb ictal motor automatisms and right upper limb dystonic posture, followed by loss of contact, cyanosis and oxygen desaturation with ictal EEG discharge on the left frontotemporal lobe, spreading to the right lobe.

Key words for video research on www.epilepticdisorders.com

Phenomenology: autonomic seizure
Localization: temporal lobe (left)
Syndrome: reflex epilepsy
Aetiology: idiopathic

TEST YOURSELF

(1) A one-year-old boy experienced recurrent episodes of unresponsiveness and cyanosis while bathing in lukewarm water. Medical history and family history were unremarkable. Clinical examination was normal. What are the possible causes of his symptoms?

A. Syncope
B. Water-induced seizures
C. Paroxysmal extreme pain disorder
D. All of these disorders

(2) What are the most common features of video-EEG in bathing epilepsy?

A. Abnormal background activity
B. Usually normal interictal EEG
C. Ictal EEG with slow high-amplitude waves mainly involving temporal regions
D. B + C

(3) What are the most common treatment measures in bathing epilepsy?

A. Avoiding the trigger of bathing
B. Surgical treatment
C. Antiseizure medications
D. A + C

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com, under the section "The EpiCentre".