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Case report

Hot water epilepsy occurring at temperature below the core temperature

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Abstract

A 6-year-old girl had water reflex epilepsy occurring at lower temperature than the core temperature. Seizures episodes consisted of a loss of consciousness absence followed by left predominant hypotonia with right fronto-temporal high voltage slow waves on the ictal-EEG. Seizures were only observed when the water was poured on scalp or face. Neuropsychological evaluation showed frontal dysfunction (Rey's figure). MRI study was normal. Oxcarbazepine permitted the disappearance of seizures and an improvement of executive disorders. In this case, the pathophysiological mechanism cannot be a hyperthermic related event. The temperature control as treatment of hot-water epilepsy could be used after the exploration of its implication in seizure induction.

Keywords: Reflex epilepsy; Hot-water epilepsy; Ictal-EEG

Epilepsy precipitated by bathing with hot water pouring over the head is known as hot-water epilepsy. A large study was published in 1988 by Satishchandra P et al. [1]. In the classical adulthood description, the temperature of water used for bathing ranges between 40 and 50 °C (customary practice in south Indians). The seizures are complex partial with or without generalization, last 30 s–3 min and can manifest either at the beginning or at the end of the bath [2]. In infancy, the seizure was induced by immersion [3]. We report a 6-year-old girl with water reflex epilepsy with right fronto-temporal symptoms in clinical, neuropsychological and electrophysiological studies. The temperature of the water inducing seizures is below the core temperature. We discuss the pathophysiological mechanisms.

1. Case report

A girl without personal or familial medical history started epilepsy at the age of two years. She never experimented febrile seizure. All the episodes had a same semiology and appeared during bath. Only one episode occurred in the swimming pool. They were complex partial seizures followed by a left predominant hypotonia during 2–3 min. One or two episodes per month occurred since the start of the disease at the age of 2 years. Only 2 episodes were noted just after the end of the bath. Her mother bathed her by pouring water with a shower. Seizures appeared only when the water was poured over her head and/or her face. No seizure was observed at immersion. The temperature of the water was always around 33 °C.

At age of 6 years, she was referred to our department for evaluation and diagnosis. Clinical examination was normal. Interictal electroencephalogram, electrocardiogram and orthostatic hypotension research were normal. We tried to induce seizure with various triggers: scalp stimulation, water with different temperatures (30–38 °C), water over the head or another part of the body with or without surprise. Seizures occurred only when the water flowed on the scalp and/or on the face. Video-EEG was realized during a bath and permitted a seizure record (Figs. 1 and 2). The temperature of water was 33 °C during this record (controlled by thermometer). Clinical presentation consisted of progressive loss of consciousness followed by left predominant hypotonia. Fear was noted before the beginning of the absence. Ictal-EEG showed a generalized

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Fig. 1. Picture from the video-EEG when the left hypotonia was noted. This picture has been taken from the video-EEG at 9:16 AM. The record started at 9:14 AM.

high voltage slow waves discharge with a predominance on the right fronto-parietal regions. Seizure duration was 16 s. Neuropsychological evaluation showed a normal intellectual quotient (WISC III) and the presence of executive dysfunction observed in Rey's Figure (Fig. 3A). There was not consequences for the schooling. MRI study was normal. Oxcarbazepine was administered (30 mg/kg/day). After one year, the evolution was characterized by the disappearance of seizure and an improvement of the executive functions (Fig. 3B).

2. Discussion

The seizures precipitated by sensory stimulus described as 'reflex or sensory epilepsy' are interesting, not as collector's item but also as providing extremely important information regarding pathogenesis of epilepsy in general and reflex epilepsy in particular. This case can be considered as a reflex epilepsy. But the features observed seem to be different from the classical Hot-Water Epilepsy [1,2,4–6]. In our patient, seizures appear to be induced by pouring of water at a temperature lower than the core temperature.

In our knowledge, only two patients were reported with water reflex epilepsy triggered by water, irrespective of its temperature [7,8]. The first patient suffered febrile convulsions in childhood, started his epilepsy at the age of 23 years and had seizures suggestive of a mesial temporal lobe origin. No imaging was realized searching for a hippocampal sclerosis [7]. The second patient was a 4-year-old girl who, at the age of 6 months, experienced complex partial seizures triggered by bathing in hot water (34–35 °C) [8]. Brain MRI detected a left parietal focal cortical dysplasia. The authors suggested that the sensory cortex might be involved in triggering seizures precipitated by a bath in hot water.

The role of water temperature was considered essential for the genesis and the treatment of this epileptic syndrome. Experimental studies on rats demonstrated that repeated hot-water stimuli (45 °C) on the head induced progressive increase in convulsive responses to stimulation, which was comparable to kindling phenomenon observed by repeated electrical stimulation [9]. Satishchandra et al. [1] postulated that the phenomenon of hyperthermic kindling might be responsible for the development of hot water epilepsy in humans. In the infancy form of hot-water epilepsy, reducing the temperature of water alone may control seizure in some patients, while antiepileptic drugs are needed in others [1,3]. From a pathophysiological point of view, combination of factors appears to be necessary in our patient to induce seizure. These factors are water as a stimulus and the head and/or the face as a stimulation area. Surprise or startling element, water on other parts of the body or sensitive stimulation on the scalp or on the face did not induce seizure. Water temperature has no influence. A hyperthermic-related event could also be excluded as a trigger. Considering the kindling phenomenon induced by hyperthermia, there is no data suggesting that kindling phenomenon exist in humans.

In pediatric studies, it was suggested that a decrease of the water temperature could be an effective treatment [3,5]. Ioos et al., suggested to decrease the temperature to 33 °C [3]. In our patient, the usual temperature used for the bath was 33 °C.

To the best of our knowledge, we first reported a neuropsychological involvement with executive dysfunction shown by Rey's figure in this type of reflex epilepsy. Hot Water Epilepsy reflex is known to be benign and self-limited. It seems that neuropsychological consequences might be observed and should be studied in-patient with Hot Water Epilepsy. It is clearly showed that cognitive functions are frequently involved in children with epilepsy. There is no specific neuropsychological pattern in pediatric epilepsy. Attention problem is frequently reported [10]. In our patient, the executive dysfunction could be related to the epilepsy by itself. On the other hand, deficits in planning and excecutive functions have been well described in children with frontal lobe epilepsy [11]. In our patient, the ictal EEG showed

Fig. 2. (A) Ictal-EEG (first part). Clinical observations following the seizure were noted on the figure by arrows: (1): she said: 'There is lots of water on the mirror' (62 s after the mother started to pour the water over her head); (2): she pushed the shower; (3): she breathed in; (4): loss of consciousness (74.5 s after the mother started to pour the water over her head). Ictal-EEG showed a generalized slow waves discharge with a predominance on the right fronto-parietal regions. (B) Ictal-EEG (second part). Clinical observation consisted of the appearance of hypotonia at the end of the seizure (arrow; 87.5 s after the mother started to pour the water over her head). Ictal-EEG showed a generalized spike-wave discharge with predominant on the right fronto-parietal regions. The consciousness recovery was noted 96 s after the mother started to pour the water over her head. The muscular tone normalized in about 4 min.