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Case report

Stimulus-induced rhythmic, periodic, or ictal discharges – A case report

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1. Introduction

Stimulus-induced rhythmic, periodic, or ictal discharges (SIRPIDs) were first described by Hirsch et al. [1] and defined as periodic, rhythmic, or ictal-appearing discharges consistently induced by alerting stimuli such as auditory stimuli, sternal rub, examination, suctioning, turning, and other patient-care activities. The authors hypothesized that the pathophysiology of SIRPIDs probably involves dysregulation of subcortico-cortical projections, particularly thalamocortical circuits, in a markedly abnormal brain with a hyperexcitable cortex. Various types of stimulation provide afferent input into the thalamus via sensory or arousal pathways that may activate these abnormal circuits [1]. However, the relationship between clinical seizures and SIRPIDs is unclear [1], although some association between SIRPIDs and clinical seizures has been observed [2,3]. Herein, we report the case of a patient with diabetes mellitus and transient hypoglycemia who exhibited SIRPIDs after correction of the hypoglycemia, which resolved spontaneously over time.

2. Case report

An 86-year-old man with a history of seizure disorder (cryptogenic generalized epilepsy with generalized tonic–clonic convulsive seizures [GTCSs]) and diabetes mellitus type 2 was admitted to the emergency department due to sudden-onset consciousness disturbance that had persisted for about 2 h. Hypoglycemia (26 mg/dL) was observed upon admission. However, after immediate correction of blood sugar levels, the patient remained stuporous (Glasgow Coma Score: E2M4V2), but no clinical GTCSs (his habitual seizures) were identified. Lumbar puncture was performed, but the cerebrospinal fluid analysis was unremarkable. Magnetic resonance imaging of the brain 4 days after admission revealed mild generalized atrophy associated with the aging process (periventricular white matter lesions). At that time, we suspected non-convulsive status epilepticus (NCSE); the video-electroencephalogram (EEG) revealed a diffuse background of slow activity (4–6 Hz) with some episodes of quasi-periodic sharp-contoured activity, at an interval of 0.8–1 s and posterior predominance lasting 2–3 s (Fig. 1). Immediately after stimulation (noxious stimulation via pinching of 1 of the patient's nipples), the quasi-periodic high-voltage activity became more prominent, lasting from 4 s (Fig. 2, 1) to 2 min (Fig. 2, 2 and 3); this activity appeared to be stimulus duration-dependent and self-limited, but not time-locked with the stimulus. Clinically, no visible convulsions could be discerned. However, NCSE was still suspected, and phenytoin and midazolam intravenous infusion was initiated, but

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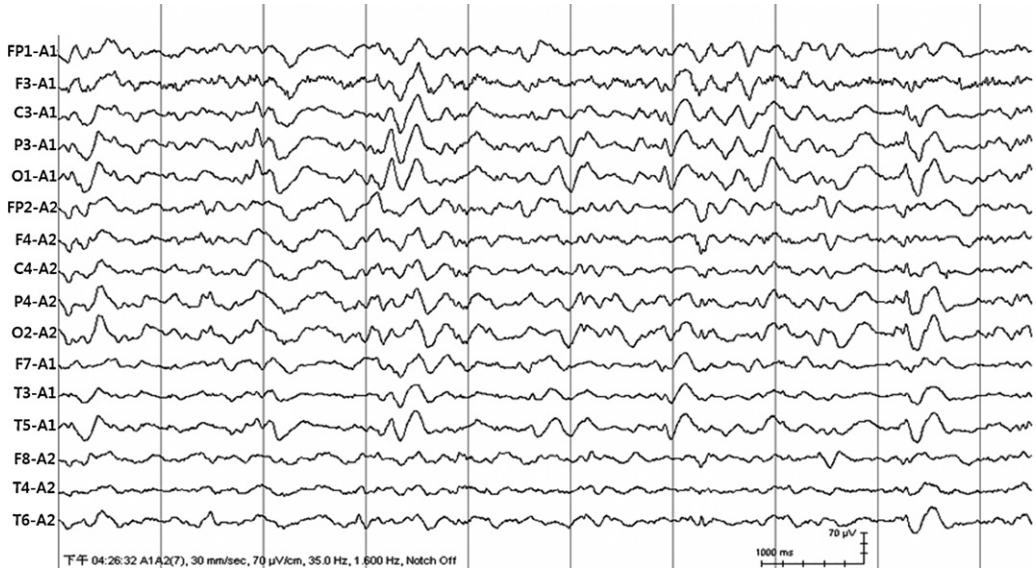

Fig. 1. Video-electroencephalography revealed a diffuse background of slow activity (4–6 Hz, not shown here) with some episodes of quasi-periodic sharp-contoured activity, at an interval of 0.8–1 s and posterior predominance lasting 2–3 s (A1–A2, 30 mm/s, 70 μV/cm, 35.0 Hz, 1.600 Hz, Notch off).

no significant changes could be identified. SIRPIDs were thus suspected. The patient showed occasional involuntary movement of the jaw resembling chewing or tremor and hand shaking, but no associated EEG findings. Within 1 week, he gradually recovered consciousness, and the suspected SIRPIDs, as well as the spontaneous intermittent quasi-periodic pattern, on the EEG disappeared. He was discharged. From the 3-month follow-up to date, the patient lived a normal daily life and had returned to his previous neurologic function.

3. Discussion

The EEG of our patient revealed generalized periodic epileptiform discharges (GPEDs) other than those associated with his habitual seizure during acute diffuse encephalopathy (hypoglycemia), and these discharges were enhanced, but not induced, exclusively by noxious stimuli, with the amplitude increasing following a pinch and decreasing after cessation of the pinch. These GPEDs were stimulus duration- and stimulus intensity-dependent and self-limited, but not time-locked with the stimulus. Thus, these discharges may fulfill the criteria for SIRPIDs proposed by Hirsch et al. [1]. The SIRPIDs appeared transiently after a hypoglycemic episode, even after blood sugar levels had been corrected, when the patient was in a stuporous state.

Murphy [4] reported a case of suspected brainstem infarction or possible embolic event, SIRPIDs determined at the first EEG during the patient's intensive care unit stay; the following EEG displayed more responsive and reactive brain activity. Although the prognosis of the patient was not discussed, it seems from these results that the patient's brain was recovering from the injury at the following EEGs. Rossetti and Dunand [3] reported a case of Creutzfeldt–Jakob disease (CJD) evolving from NCSE through SIRPIDs to generalized periodic discharges. They argued that SIRPIDs in CJD could be related to seizure occurrence, rather than being an epiphenomenon. Fluri et al. [2] reported a case of severely hypoxic encephalopathy, with a prolonged series of bilateral periocular myocloni with concomitant evolving seizure activity in the EEG (SIRPIDs), elicited exclusively by touching the area innervated by the ophthalmic nerve. In addition, the patient also experienced prolonged generalized seizures following stimulation, instead of brief myoclonic jerks (clinical bilateral periocular twitching). The authors concluded that SIRPIDs can be induced by exteroceptive stimuli exclusively applied to a distinct body region.

Although the relationship between clinical seizures and SIRPIDs is unclear [1], Rossetti and Dunand [3] and Fluri et al. [2] have described an association between SIRPIDs and clinical seizures. SIRPIDs may be observed in a broad range of underlying conditions, including as an intermediate form reflecting marked cortical insult [3]. As proposed by Hirsch et al. [1], SIRPIDs may be considered a type of reflex epilepsy (NCSE). Wolf [5] suggested that the mechanism of reflex epilepsy may depend on the activity of a function-related network of both established and plastic links between brain regions that are modified by the effects of factors related to the specific stimulation. In SIRPIDs, exteroceptive stimuli may result in the recruitment of established links of the function-related network of the abnormal brain with a hyperexcitable cortex. The state of hyperexcitability and the linking of the function-related network are not static in SIRPIDs. Thus, the phenomenon is usually transient and not persistent, as in our patient and most previous reports. However, the relationship between SIRPIDs and the underlying disorder, the state of hyperexcitability of the cortex, the exteroceptive stimulus, and the patient's prognosis is still unclear and needs to be further elucidated.

Additionally, the boundaries between NCSE and encephalopathy may be imprecise and vague. The category of generalized discharges also includes triphasic waves (TWs). TWs are periodic and generalized, typically frontally predominant with a frontal-occipital lag. Because TWs have generally not been considered epileptiform, they are often not included in the GPED categorization. However, the typical EEG response after alerting stimuli in patients with TWs is still being debated. Hirsch et al. also suggested that TWs may be good models for studying SIRPIDs [1]. Our patient's EEG revealed "triphasic-like waves" without clear frontal-occipital lag, which did not respond to IVA of BZD. Thus, these triphasic-like waves were classified as GPEDs. However, the differentiation between borderline-NCSE and TW toxic encephalopathies depends on the clinician's point of view. Thus, further research is necessary to explore the relationship between TWs, SIRPIDs, and NCSE.