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Protracted continual cyclical catatonia-like ictal episodes in a patient

Tzu-Hsin Huang[1]; Tsang-Shan Chen[2]; Chin-Wei Huang[1]

[1]Department of Neurology, National Cheng Kung University Hospital, College of Medicine, National Cheng Kung University, Tainan, Taiwan

[2]Department of Neurology, Tainan Sin-Lau Hospital, Tainan, Taiwan

***Corresponding Author(s): Chin-Wei Huang**

Department of Neurology, National Cheng Kung University Hospital, College of Medicine, National Cheng Kung University, Tainan, Taiwan

Tel: 886-6-2353535/5485, Fax: 886-6-2374285

Email: huangcw@mail.ncku.edu.tw

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Abstract

Catatonia and Nonconvulsive status epilepticus (NCSE) may share clinical features and it may be difficult to distinguish between them simply based on the clinical manifestation and neurological examination. Catatonia, associated with numerous general medical and neurological conditions, could be the presentation of an epileptic seizure and it could be difficult to differentiate catatonia from NCSE. Here, we report a unique case of NCSE presenting as unusual continual, cyclical catatonia-like ictal episodes.

Introduction

Catatonia is an abnormal neuropsychiatric condition that affects both behaviors and motor function, and results in unresponsiveness in someone who otherwise appears to be awake [1]. It is a clinical challenge to differentiate catatonia from other neuropsychiatric conditions. Catatonia could be the presentation of an epileptic seizure and it could be difficult to differentiate catatonia from Nonconvulsive status epilepticus (NCSE). Here, we report a case of NCSE presenting as continual, cyclical catatonia-like ictal episodes.

Case Report

A 64-year-old female, diagnosed as primary Sjogren syndrome with bilateral retinopathy and cataract for more than 20 years, was admitted to our hospital for her cyclical catatonia-like appearance, dull response and amnesia episodes, which recurred consecutive 24 hours every three days for two years, making her become near totally bed-ridden state.

Three years ago, she started having episodes of headache, dizziness, and slowness in gait, lasting for several hours. Simi-

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lar episodes recurred every three days since that time. One year later, she had two times of generalized tonic clonic seizure (GTC). Antiepileptic drugs (levetiracetam 500 mg twice per day, and topiramate 50 mg twice per day) were administered and there was no GTC recurrence thereafter. Brain MRI revealed some old infarcts in left pons and left corona radiata. EEG did not show remarkable findings.

Nevertheless, the episodes progressed with persistent upward staring, mouth opening and without responses to external stimulation two years ago. She was then admitted to our ward for continuous video EEG monitoring (Figure 1a). Her Cerebrospinal fluid (CSF) study was normal. During catatonia phase, EEG revealed continuous, synchronous, medium voltage spikes with frequency of 10.5Hz in bilateral frontotemporal area.

We administered intravenous midazolam infusion to ameliorate the epileptic seizure activity. EEG showed rapid transition from continuous spikes activities to normal activities at the end of midazolam infusion (Figure 1b).

Another trial in the intensive care unit to reduce and clarify the presence of muscle artifacts was carried out, and there were no significant difference in EEG before and after the administration of succinylcholine (Figure 1c & 1d). We then adjusted antiepileptic drugs (AEDs) to topiramate (100 mg twice per day), levetiracetam (1000mg twice per day), valproic acid (250mg twice per day), perampanel (6 mg before sleep). She gradually improved and had no catatonic-like episodes 3 months later, and she could walk by herself at the eighth month's follow-up.

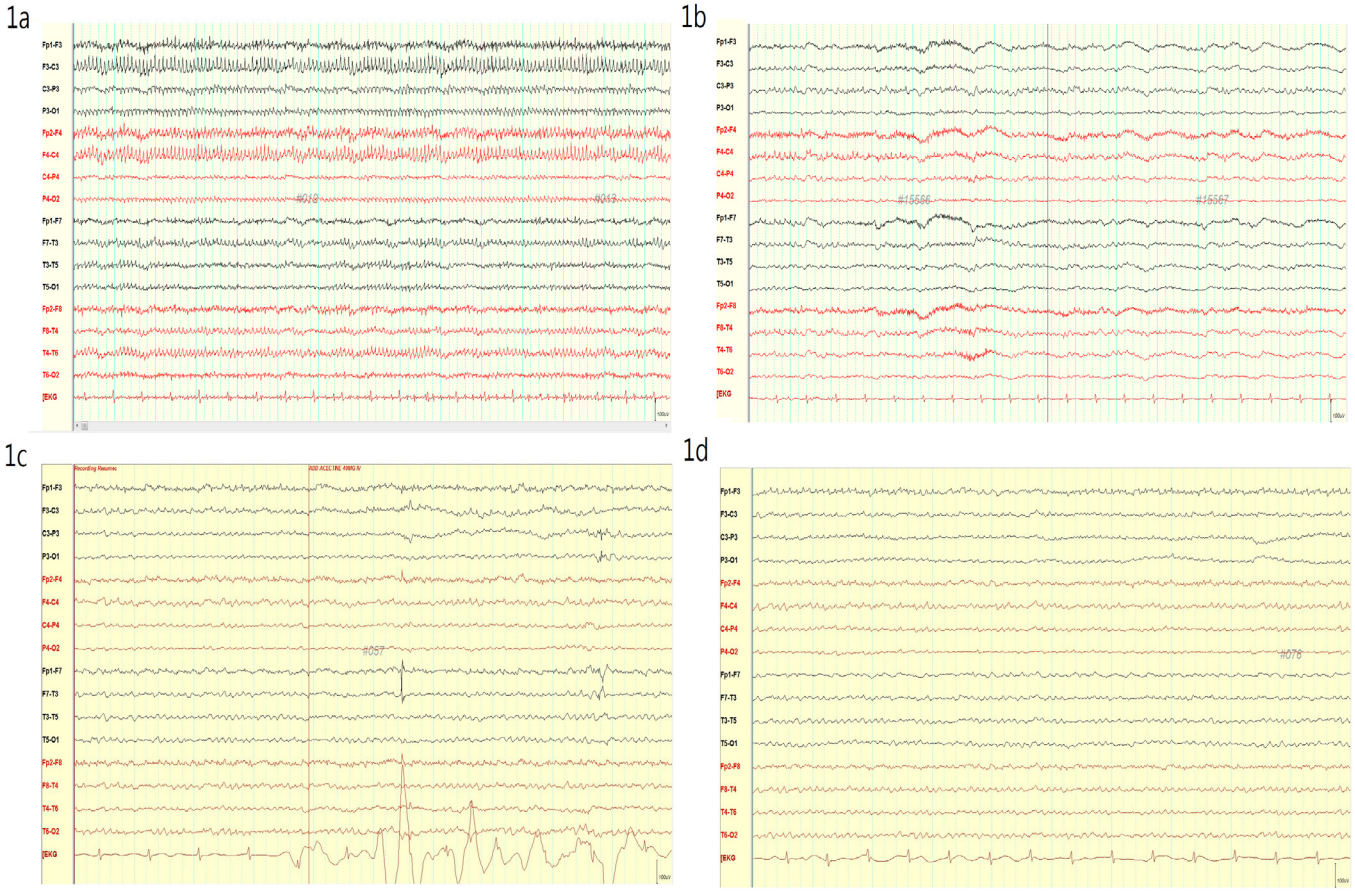

Figure 1: **(1a)** EEG during catatonia-like phase showed continuous, synchronous, medium voltage spikes with frequency of 10.5Hz in bilateral frontotemporal area. **(1b)** EEG after infusion of midazolam showed rapid transition from continuous spikes activities to normal activities. EEG before **(1c)** and after **(1d)** succinylcholine infusion, showed no significant difference in EEG recordings.

Discussion

Catatonia is defined as the presence of three or more of the following criteria in DSM-V: catalepsy, waxy flexibility, stupor, agitation, mutism, negativism, posturing, mannerisms, stereotypies, grimacing, echolalia and echopraxia [1]. Periodic catatonia is a rare condition and is usually associated with schizophrenia [2]. Although our case appeared to match the criteria with stupor, mutism and negativism, we did not find any underlying history of psychiatric disorder or antipsychotics exposure. Our investigation on this case supported the diagnosis of NCSE.

NCSE may produce an alteration of consciousness and behavior, rigidity, mutism, catatonic posturing and autonomic signs [3]. These changes may occur unaccompanied by obvious convulsive seizures or a history of epilepsy. To confirm the catatonia-feature in our patient is an ictal manifestation, we conducted both midazolam and succinylcholine trials. The epileptiform discharges were effectively reduced by midazolam, while persisted with succinylcholine infusion. The EEG findings also matched the criteria of SE, with either generalized, symmetrical, typical spike-and waves with 3-3.5 Hz, or asymmetrical repetitive complexes of two or more spikes and atypical spikes and slow waves.

Reviewing the literature, most of cyclical pattern of seizures were described in patient dwelling in intensive care unit, a study which collected 13 critically ill patients of all ages, most of them were non-convulsive [4]. The cyclical pattern of the catatonia-

like NCSE features the unique chronicity of the epileptic network in our case.

We are not in favored of autoimmune etiology as the basic normal autoimmune profile, patient's age and its relatively long disease course. In addition, CSF study was normal, and survey for metabolic and hereditary origin was unremarkable as well. Further survey on rare auto-antibodies, such as anti-Iglon5, and paraneoplastic antibodies might be indicated.

Some studies suggest the involvement of dysfunction of projection between the basal ganglia with the cortex and thalamus in catatonia [1]. Although the precise mechanism is currently unknown, the cyclical clinical features probably root in the dynamic balance between inhibitory and excitatory mechanism. In our case, although the etiology is unknown, the epileptiform discharges related frontal and temporal dysfunction and/or probable hypoperfusion may have contributed to this clinical phenomenon [5]. Further studies on active inhibitory mechanisms, neuromodulators, and the activity of hyperpolarizing ion pumps and changes in pH are warranted.

Conclusions

We report a rare and unique case of NCSE presenting with cyclical catatonia-like ictal episodes, who was well controlled with AED polytherapy. Video EEG monitoring, with the aid of intravenous benzodiazepine and succinylcholine, is important in confirming the diagnosis of NCSE and differentiation of catatonia.

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