Case Report

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Recurrent Syncope Triggered by Temporal Lobe Epilepsy: Ictal Bradycardia Syndrome

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Ictal asystole is potentially lethal, and known to originate from the involvement of limbic autonomic regions. Appropriate treatment must include an antiepileptic drug and the implantation of a pacemaker. We report the case of a 54-year-old male with recurrent syncope secondary to ictal asystole triggered by temporal lobe epilepsy. This was confirmed by combined Holter and video-electroencephalogram monitoring. (Korean Circ J 2012;42:349–351)

KEY WORDS: Bradycardia; Syncope; Electrocardiography.

Introduction

Syncope is defined as a transient loss of consciousness, usually leading to falling, which results from transient inadequate cerebral perfusion. The most common causes of syncope are reflex-mediated syncope and orthostatic hypotension. Rarely, cardiac bradyar-rhythmias induced by seizures can give rise to syncope. Ictal bradycardia syndrome is a rare cause of syncope but can be a potentially life-threatening syndrome associated with partial epilepsy. It has been implicated as a cardiac cause of sudden unexplained death in epilepsy (SUDEP).

Cardiac pacemaker implantation along with antiepileptic drug therapy may be necessary to minimize the possibility of death. We report a patient with recurrent syncope secondary to ictal asystole triggered by temporal lobe epilepsy.

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Case

A 54-year-old Korean Canadian man was admitted due to the worsening of a recurrent syncope during the past two years. He had experienced syncope once or twice a year, especially during or after eating. He had been treated with carbamazepine at a local clinic. He reported symptoms of jamais vu, dizziness, and a syncope lasting less than a minute, followed by post-ictal confusion for 30 minutes. There was no identified trigger, but the attack was usually developed while eating. He did not smoke cigarettes or drink alcohol and had none of the classic cardiovascular risk factors. His blood pressure was 138/92 mm Hg, respiratory rate was 20/min, and heart rate was 91 beats/min. A physical examination revealed no specific findings. The magnetic resonance imaging result was normal. He underwent a video-electroencephalogram (EEG) monitoring combined with a Holter monitoring.

The first episode of vacant staring developed while he was eating lunch. His head dropped and showed subtle generalized convulsive movements for a few seconds. EEG showed generalized rhythmic theta activity at that event (Fig. 1). A severe sinus bradycardia and asystole started 22 seconds after seizure onset and lasted 40 seconds (Figs. 1 and 2). Three episodes of seizure attack and a long sinus pause developed during 24 hours of EEG and Holter monitoring (Fig. 2).

On the basis of combined Holter and video-EEG monitoring, the diagnosis of ictal bradycardia syndrome was confirmed. We recommended permanent pacemaker implantation, but he wanted the implant in his country. Therefore, he was discharged on medical thera-

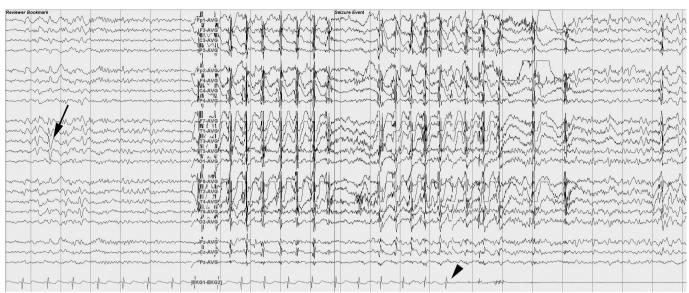


Fig. 1. Electroencephalogram and electrocardiogram. Electroencephalogram showed rhythmic theta activity and sharp waves in the T3 area (arrow) at first, and then generalized rhythmic theta activity. Asystole started 22 seconds after seizure attack, as shown in the electrocardiogram channel (arrow head).

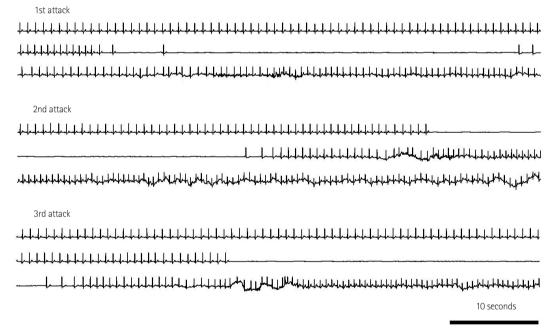


Fig. 2. Holter recording. Holter recording showed three episodes of a long sinus pause of 40 seconds, followed by junctional escape rhythm, and then sinus tachycardia.

py for epilepsy. Later, he had a pacemaker implanted in Canada and did not experience recurrence of syncope.

Discussion

Cardiac rhythm changes occur in a majority of epileptic seizures. The most common cardiac arrhythmia observed during epileptic seizures is sinus tachycardia, which occurs in >90% of seizures and is usually of no consequence.⁴⁾⁵⁾ Marked sinus bradycardia or asystole is rarely associated with seizures. Patients with bradycardia and asystole during an epileptic seizure are classified as having ictal bradycardia syndrome.²⁾ Ictal bradycardia syndrome is a rare feature of patients with focal epilepsy mostly from the temporal lobes and has been implicated as a cardiac cause of SUDEP.3) Ictal and interictal epileptogenic activity spreading from the temporal lobe areas may interfere with cardiovascular modulation in neighboring structures of central autonomic control, such as the amygdale, the insular or orbitofrontal cortex, the cingular gyrus and their pathways. ⁶⁾⁷⁾ Electrocardiogram (ECG)-triggered microstimulation of the left posterior insular cortex in rats may lead to complete heart block



and asystolic death. Electroconvulsive therapy in humans can trigger asystole simultaneous with the onset of the electrical stimulus, both suggesting a direct mechanism on the heart.

Park et al.9) reported a case of frontal lobe brain tumor related to seizure and complete atrioventricular block, but comcomitant Holter and EEG recording was not conducted during seizure events. In our case, combined Holter and video-EEG monitoring could confirm the origin of seizure and diagnosis of ictal bradycardia syndrome simultaneously. Concomitant Holter and EEG recording should be considered when patients with temporal lobe epilepsy develop recurrent syncope.

There are no guidelines for the management of patients with ictal arrhythmia. One prospective study⁸⁾ using implantable loop recorders to get ECG in patients during typical seizures showed that 4 out of 19 patients had bradycardia or periods of asystole. Three out of these four had potentially fatal asystoles. These authors concluded that the clinical characteristics of patients with periictal cardiac abnormalities are very similar to those at greatest risk of SUDEP. One case reported that implantation of a cardiac pacemaker while continuing antiepileptic drugs rendered a patient free from ictal symptoms and prevented ictal syncope and subsequent trauma.

Our case is a patient with recurrent, unexplained syncope for several years, with no personal or family history of epilepsy. The patient suffered symptoms such as jamais vu, which were suggestive of partial seizures. The attack was usually developed while eating, but it was different from situational syncope such as swallowing syncope that is caused by a reflex of the involuntary nervous system called the vasovagal reaction. 10) To rule out situational syncope, we performed EEG analysis, which showed rhythmic theta activity and sharp waves in the left temporal area and generalized rhythmic theta activity. Asystole started a few seconds after seizure onset. He was discharged on medical therapy for seizure and later implanted with a pacemaker.

In conclusion, ictal bradycardia syndrome should be postulated when patients with temporal lobe epilepsy develop recurrent syncope. The diagnosis can only be confirmed by concomitant Holter and EEG recording during an event. The double headed treatment allows an adequate, comprehensive prevention of potentially deleterious complications.

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