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Case Report

Presence of both Complex Partial Seizures and Atrio-Ventricular Block

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Abstract

Cases in which epilepsy and atrio-ventricular (AV) block coexist are unusual. A 31-year-old man presented with syncope. Holter ECG showed the second-degree AV block and the longest cardiac pause was 6.3 seconds. Even after receiving pacemaker, he repeated consciousness cloudiness. Electroencephalogram revealed spike and waves on the left temporal region, suggesting complicated with complex partial seizure. There was no pathogenic mutation in KCNA1 encoding Kv1.1 potassiume channel of which deficient mice displayed both epilepsy and AV block. Epileptic discharge in the left temporal region could result in bradycardia and sometimes it might be a cause of sudden unexpected death in epilepsy. Neurologists and cardiologists should pay attention to this rare disorder.

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- KCNA1

INTRODUCTION

There have been rare cases showing epilepsy with arrhythmia [1]. Arrhythmia is thought to be the major cause of sudden unexpected death in epilepsy and such epilepsy may be called as arrhythmogenic epilepsy. In some of these rare epilepsies with arrhythmia, arrhythmogenic channel abnormalities were discovered. Long QT syndrome is one of them. In this syndrome, lethal ventricular arrhythmia occurs [2,3]. On the other hand, epilepsy complicated with atrio-ventricular (AV) block has been reported [4]. We describe a case of complex partial seizure with AV block. He underwent permanent cardiac pacemaker and was treated with anti-epileptic drugs and thereafter he did not experienced syncope or seizure.

CASE PRESENTATION

A 31-year-old right-handed man presented with syncope. He first experienced syncope in the bathtub. He was transferred to another hospital. Although Electroencephalogram (EEG) did not show epileptic discharge, he was diagnosed as epilepsy and treated with 400 mg/day valproate. But he discontinued the medication after two weeks. The second attack was in the dining room. After the second attack, he came to our hospital. He was alert and normal on neurological examination. There was no past history of head trauma or neurological disease. Also, there was no family member suffering from epilepsy or cardiac disease. His parents were non-consanguineous. Although Electrocardiogram (ECG) was normal and QTc was 0.367, Holter ECG revealed the second-degree AV block and the longest cardiac pause was 6.3 seconds (Figure 1). He was told to go to the department of cardiology in the other hospital immediately and further tests of brain magnetic resonance imaging and EEG were cancelled. The level of BNP was 4.2pg/mL. He was not diabetic or hypothyroidism. Ejection fraction calculated from echocardiogram was 73%. Cardiac angiography was normal. He underwent a DDDR permanent cardiac pacemaker. Even after receiving pacemaker, he experienced consciousness cloudiness several times. During a football game, he stood alone without falling in the ground. In the other situation, he did not respond to his wife's answer for a few seconds in the kitchen. He has never experienced a generalized convulsion. He came to our hospital again. His pacemaker worked correctly. As complex partial seizure was suspected, regular 30-min EEG was recorded. The background EEG was 11-12 c/s alpha waves. There was no change during photo-stimulation and hyperventilation. The EEG revealed spike and wave complex predominantly on the left temporal region (Figure 2). He was diagnosed as having complex partial seizure as well as the second-degree AV block. Counts of blood cells, liver function, renal function, and electrolytes including magnesium were all normal. The levels of serum lactate, pyruvate, and angiotensin converting enzyme were also normal. Brain lesion was not found in computed tomography. There was no neuropathic change in his nerve conduction study. His epileptic attack was stopped by combination therapy with valproate (800 mg/day) and carbamazepine (400 mg/day). Since his complex partial seizure was controlled, the pacing function of the pacemaker has not worked for longer than three years. Derived informed consent for the gene diagnosis,

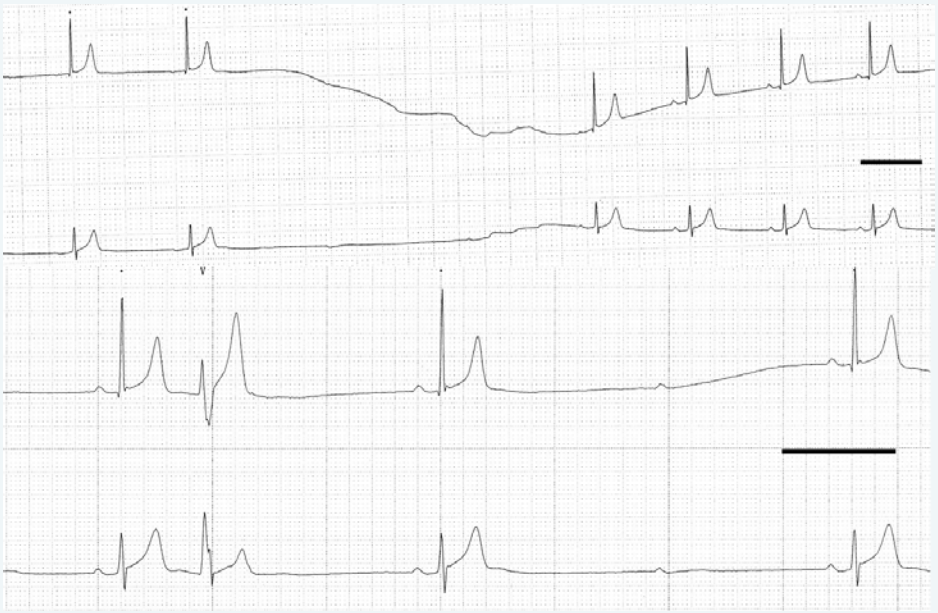

Figure 1 Result of Holter ECG. The longest cardiac pause was 6.3 seconds (upper). The second-degree AV block was found (lower). The lines indicate one second.

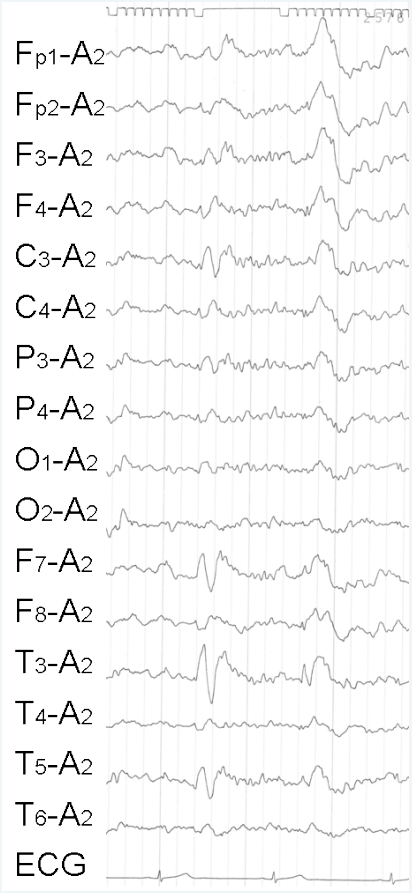

Figure 2 Electroencephalogram revealed spike and wave complex predominantly on the left hemisphere. As the routine recording showed activation of the A1, all electric activities were automatically re-calculated with the reference electrode of A2. The maximum point of the epileptic discharges was located at T3.

KCNA1 encoding Kv1.1 potassium channel was examined, since mice lacking Kv1.1 displayed interictal cardiac abnormalities including a fivefold increase in AV block [5]. Although we found two heterozygous (684T>C, 804G>C) and one homozygous non-synonymous polymorphism (1440T>A), there was no pathogenic mutation.

DISCUSSION

There have been a few reports showing that AV block was complicated with epilepsy [4]. Cardiac arrhythmia including AV block is considered to be associated with Sudden Unexpected Death in Epilepsy (SUDEP).

The presented case showed no brain lesion or cardiovascular lesion. Moreover, he has no clinical feature suggesting mitochondrial diseases, vasculitis, or granulomatous diseases. These facts indicated that his epilepsy and AV block could not be secondary but primary events and could be associated with each other. The fact that antiepileptic drug prevented AV block for three years indicates that his epilepsy could be an arrhythmogenic epilepsy. Oppenheimer et al. reported that the possible relationship between insular epileptic discharge and cardiac arrhythmia. The electric stimulation of the right insula caused tachycardia and elevation of blood pressure. On the other hand, the left insular stimulation resulted in bradycardia and decrease of blood pressure [6]. Our case supported their hypothesis.

Treatment of arrhythmogenic epilepsy might be complex. Antiepileptic drugs can cause AV block by themselves. In cases with syncope, it is safer that installation of permanent pacemaker is preceded. Afterwards antiepileptic drugs should be added. Even after epilepsy is controlled like our patient, permanent pacemaker could be necessary to prevent SUDEP.

In summary, we described a sporadic case with complex partial seizure complicated with second-degree AV block

which needed a permanent cardiac pacemaker. Although arrhythmogenic epilepsy is unusual, a full diagnostic workup including EEG and Holter ECG is important even in cases either of which is obviously abnormal.

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