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Clinical letter

Cardiogenic paroxysmal autonomic events mimicking complex partial seizures

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1. Introduction

A cardiogenic paroxysmal autonomic event (CPAE), including swallow syncope, can be mistaken for an epileptic seizure. In the case of a CPAE, an underlying cardiac pathology should rapidly be considered as delayed treatment of the underlying cardiac condition poses a significant mortality risk, including the risk of sudden cardiac death (SCD). Further, when CPAE is misdiagnosed as an epileptic seizure and is treated by enrollment in antiepileptic drug (AED) trials, cardiac bradyarrhythmia can be aggravated by some ion channel active AEDs; because of this, care must be taken, as a hasty diagnosis of epilepsy can have significant psychosocial consequences.

Swallow syncope is a rare form of syncope that causes inhibition of cardiac conduction. It is a dysautonomic syndrome associated with hypersensitive vagal activation due to esophageal stimulation. Swallow syncope, through excessive parasympathetic stimulation and sympathetic inhibition, can induce a variety of bradyarrhythmias. In this article, we report the case of a patient with a swallowing-induced AV block who was initially misdiagnosed as suffering from medically intractable complex partial seizures.

2. Case report

A 60-year-old man was admitted to the hospital with a history of recurrent paroxysmal autonomic events (PAEs), which had developed 18 months prior to admission. After daily episodes of sudden epigastric rising while eating, facial flushing developed suddenly and subsided abruptly; these events were triggered by swallowing, especially when drinking cold beverages or soups. The patient usually became unresponsive during these events and sometimes lost consciousness, but always regained full consciousness immediately without confusion. His events were misinterpreted as complex partial seizures and classified as abdominal auras evolving into dialeptic seizures. Due to the initial misdiagnosis, AEDs were used as a treatment for several months but had no effect on the PAEs.

The patient had a total of six PAEs during his 3-day hospital stay for video-EEG monitoring (1 ECG and 18 EEG channels). During this time, there were no specific findings in the interictal EEG. All events occurred immediately after drinking cold water or soup. Events were reported by the patient or his wife; the patient pushed a button when he felt epigastric rising, and his wife recognized the events by his facial changes (such as initial paleness and late flushing) (Supplementary Fig. 1, Supplementary Video). The EEG showed nonspecific findings, including increased theta waves before the onset of clinical symptoms

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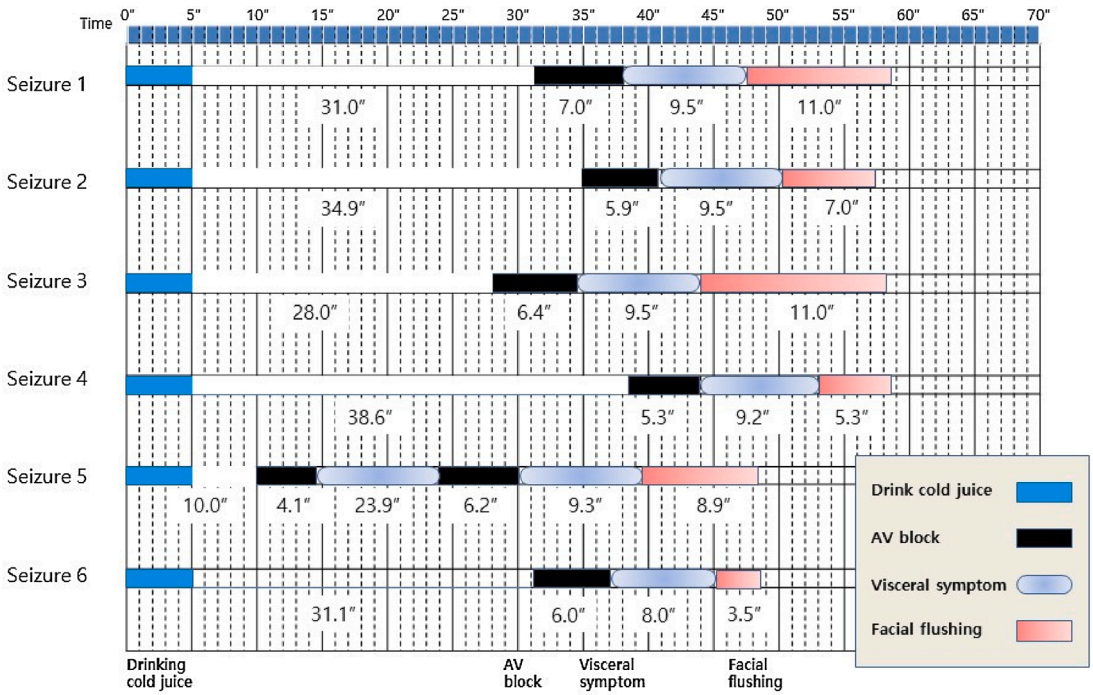

Fig. 1. Schematic drawings of clinical seizures based on time. Drinking cold juice induced AV block followed by visceral symptoms and facial flushing. Facial flushing then stopped abruptly, and the face regained a normal appearance.

Table 1
Correlation of latency period and duration of clinical manifestations. A Kendall's tau-b correlation was run to determine the relationship between the parameters among the six events. There was a positive correlation between AV block duration and PAE duration ($\tau b = 0.690$, $p < 0.1$).

	Average (S.D.)	1.	2.	3.	4.	5.
1. Latency period to AV block (sec)	28.2 (9.30)	1	−0.143	−0.411	−0.414	−0.414
2. AV block duration (sec)	5.8 (0.93)		1	0.000	0.690^{+}	0.690^{+}
3. Visceral symptom duration (sec)	11.3 (5.60)			1	0.772*	0.772*
4. Facial flushing duration (sec)	7.8 (3.07)				1	1.000**
5. PAE duration (sec)	17.0 (3.54)					1

PAE: paroxysmal autonomic event.
$^{+}$ $p < 0.1$.
* $p < 0.05$.
** $p < 0.01$, Kendall tau-b.

(Supplementary Fig. 1). The AV block was identified on the ECG, and it was noted that all of the visceral symptoms and the following facial changes developed suddenly after the AV block occurred.

To analyze the patient's symptoms, we measured the duration of the following: latency period from consuming cold beverages or soups to the occurrence of AV block; the AV block, visceral symptoms including epigastric rising; facial flushing; and the PAEs combined with visceral symptoms and facial flushing (Fig. 1). Analyzing the correlation between these parameters showed that the AV block duration was moderately positively correlated with PAE duration, especially with the duration of facial flushing ($\tau b = 0.690$, $P < 0.1$) (Table 1).

The patient's brain magnetic resonance imaging (MRI) showed several chronic ischemic lesions on both cerebral white matter, but no other abnormalities were observed. No abnormal enhancement was observed in the post-contrast image. The esophagogastroduodenoscopy revealed no abnormal findings other than chronic gastritis. In cardiac evaluation, the patient's holter monitoring verified a high degree AV block (6:1) with pauses up to 6.16 s; these findings correlated with the symptoms the patient reported. After the implantation of a dual-chamber permanent pacemaker, the PAEs disappeared.

3. Discussion

As there are many diseases that present with PAEs, caution should be exercised in making a final diagnosis. In particular, CPAEs must be differentiated from complex partial seizures, since epileptic seizures pose a well-known risk of misdiagnosis and unnecessary use of AEDs.

Swallow syncope is an unusual type of neurogenic situational syncope. The most commonly postulated mechanism is that the increased and excessive vagal reflex activation during swallowing causes cardiac inhibition [1]. During swallowing, afferent impulses from the esophageal plexus travel via the vagus nerve to the nucleus solitarius tract in the medulla oblongata. Subsequently, a corresponding signal that regulates involuntary peristalsis travels down the parasympathetic efferent fibers and through the esophageal branch of the vagus nerve [2]. The presence of reflex arcs between afferent sensory fibers and efferent parasympathetic fibers of the cardiac branch results in inappropriate vagal activation, with bradycardia, disturbance to the conduction system, and hypotension secondary to vasodilation [1,3]. The exact mechanism remains to be elucidated; however, excessive parasympathetic stimulation of the heart seems to be the central mechanism.

PAEs are related to sympathetic and parasympathetic activity. Prodromal symptoms such as sweating, facial pallor, epigastric discomfort and vague nausea, pupillary dilatation, and palpitations occur because of reduced blood flow caused by sympathetic and vasopressin-induced vasoconstriction in addition to low blood pressure. Facial flushing, one of the postictal events, occurs 2–4 s after the circulation restarts and during an overshoot in arterial pressure following the asystole [4].

In the present case, AV block always preceded the occurrence of autonomic events, and the symptoms were caused by swallowing. Because of this, the patient was diagnosed with swallow syncope. The

mechanism of syncope happens as swallowing cold beverages or soups stimulates the vagal tone, which triggers the dysfunction of the cardiac conduction system up to cardiac asystole. After a variable latency period of AV block (10.0~38.6 s), the patient experienced paroxysmal dysautonomia events, including epigastric rising and the paling of his face. After the dysautomonia was revealed, his face displayed flushing and regained normal appearance abruptly.

Considering that the latency period of AV block varied in this case, we can guess that this was a complex reflex syncope, not a simple reflex syncope. It is thought that variable latency was induced by the difference in maturation process according to the patient's condition and heart-brain connection state. The patient's PAE duration was well-correlated with his AV block duration, suggesting that the cerebral ischemic duration influenced the PAE. A PAE is the combination of visceral symptoms (failure of autonomic function) and facial flushing (recovery of autonomic function). In the case presented here, AV block duration was more strongly correlated with the duration of facial flushing than with the duration of the visceral symptoms. This suggests that, in a dysautonomic state, AV block duration might be more strongly correlated with the recovery of autonomic function than with its failure.

In conclusion, since CPAEs carry a considerable mortality and morbidity risk, we should consider cardiac pathology and evaluate cardiac function. In the present case, the patient's PAE presented similar to a complex partial seizure, but the syncopal event was found to be due to a swallowing-induced AV block. Therefore, when evaluating a patient presenting with PAEs mimicking epileptic seizures, it is important to simultaneously perform continuous ECG data recording and video-EEG monitoring to exclude cardiogenic causes and provide a proper diagnosis.

Declaration of Competing Interest

The authors report no declarations of interest.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.seizure.2020.09.003.

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