Case Report

A case of idiopathic ventricular fibrillation in which implanted loop recorder was useful in application decision for implantation of implantable cardioverter-defibrillator

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ABSTRACT

Establishing a symptom–rhythm correlation in patients with unexplained syncope is complicated because of its sporadic, infrequent, and unpredictable nature.

Recently, an implantable loop recorder (ILR) has become available to evaluate undiagnosed recurrent arrhythmic episodes particularly in unexplained syncopeces, and its usefulness has been reported in patients with syncope episodes that remain unexplained after conventional work-up.

A 65-year-old man was referred to our hospital for loss of consciousness with nocturnal paroxysmal seizures. He had experienced several similar episodes. No family history of sudden death was evident, and apparent structural heart disease was absent. Coronary angiography with intracoronary ergonovine provocation showed vasospasm in left coronary artery without organic stenosis. Ventricular tachyarrhythmias were not induced by programmed electrical stimuli. According to the guideline, he was at once categorized as having class IIb indication for implantable cardioverter defibrillator implantation. However, his symptoms were not relieved despite administration of anti-anginal medications including nitrates and calcium antagonist. Implantation of an ILR was performed and revealed an episode of ventricular fibrillation during generalized-convulsion attack during sleep. ILR is useful in determining the presence of fatal arrhythmias during syncope, although conventional diagnostic testing, such as electrocardiogram, Holter monitoring, and external loop recording, is inconclusive.

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Case Report

A 60-year-old male suffered from recurrent episodes of nocturnal paroxysmal convulsion and was admitted to our hospital. His attacks were accompanied with or without generalized convulsion since the previous three years. The frequency of attacks was twice or three times per year, mostly at mid-night hours during sleep. His 12-lead electrocardiogram (ECG) showed sinus rhythm with a heart rate of 48 bpm, a PR interval of 180 ms, a QRS duration of 100 ms, a corrected QT interval of 410 ms, and saddleback type ST-elevation with a J-wave amplitude > 2 mm in lead V2 (Fig. 1A). J wave was also observed in the inferolateral lead. After placement of the right precordial leads in superior positions, ECGs revealed coved-type ST elevation with a J-wave amplitude > 2 mm in leads V1–2 (Fig. 1B). Holter ECG monitoring revealed only frequent, unifocal ventricular premature contractions. Echocardiography, brain electroencephalography, and magnetic resonance imaging of central nervous system revealed negative findings. Coronary angiography demonstrated no significant stenotic lesions with vasospasm by ergonovine provocation test (Fig. 2). Cardiac electrophysiologic study (EPS) failed to induce ventricular tachycardia (VT) or ventricular fibrillation (VF) by up to three extra-stimuli coupling at two drive cycle lengths at right ventricular apex and outflow tract before and after isoproterenol infusion. Although this patient had undergone extensive evaluation, no arrhythmic events had been uncovered. Molecular genetic analysis was not performed. Based on these findings, the indication of implantable cardioverter defibrillator (ICD) implantation was class IIb according to the Guidelines for Non-Pharmacological Therapy of Cardiac Arrhythmias (published from Japanese Circulation Society 2011) [1], and he was treated as an out-patient. In order to clarify the exact cause of syncope, he received implantation of an implantable loop recorder (ILR). One month later following ILR implantation, his wife reported a generalized-convulsion attack during sleep. Telemetry reading of...
the ILR revealed an episode of VF (Fig. 3). In addition, a positive J-wave deflection inscribed on the S wave emerged before VF occurrence and disappeared after VF termination by the record of ILR (Fig. 4).

Subsequently, an ICD was implanted to manage the VF. To date, no more VT nor VF or any therapeutic shock was recorded by the backup ICD.

**Discussion**

In this case, although recurrence of nocturnal syncope was observed, however, VF or VT with hemodynamic compromise was not induced by EPS, so ICD implantation was not initially indicated according to the guidelines for non-pharmacotherapy of cardiac arrhythmia published by the Japanese Circulation Society with the applicability criterion of class IIb [1]. An ILR was implanted for follow-up scrutiny, and consequently ICD implantation was determined by record of spontaneous VF episode. The documentation of fatal arrhythmia by ILR offered decisive evidence for the ICD implantation.

The prognosis of Brugada syndrome (BS) [2] varies depending on reports [3–7], and there are no clear standards with regard to what kind of management should be performed in patients with applicability of class IIb for ICD implantation as in our case. To date, the decisions are entrusted to the clinician’s arbitrary judgment.

The management of patients with symptomatic BS slightly differ between Europe and the USA and Japan. In Europe and the USA, the patient is determined as being suitable for ICD implantation if they have both a coved-type Brugada ECG and history of syncope. In addition, according to the second consensus report regarding BS [8], if a patient has a spontaneous type 1 ECG (coved-type ST increase of 2 mm or more electrical potential at J point) with a history of
Fig. 3. Implanted loop recorder revealed ventricular fibrillation episodes twice: (A) 1st event (sustained for 60 s) and (B) 2nd event (sustained for 96 s).
syncope, there is ICD implant applicability of class I when extracardiac factors are excluded. In addition, patients that exhibit a type 1 ECG after the administration of a Na channel blocker are indicated for ICD implantation with applicability of class IIa if there is a history of syncope and when extracardiac factors are excluded. Therefore, the presence of a syncopal episode is emphasized regarding indication for ICD implantation in western countries. The multi-institutional research by Eckardt et al. [6] and the report by Giustetto et al. [7] demonstrated that the incidence of cardiovascular events in patients with a history of syncope was significantly high as compared to the asymptomatic cases with incidence of 6.2% and 8.6%, respectively.

In Japan, the presence of VF induction in electrophysiological test is the major clinical factor with regard to ICD implantation according to the Japanese guideline. The cardiovascular contract research by the Ministry of Health, Labor and Welfare as well as J-IVF research [9] by the Idiopathic Ventricular Fibrillation Research Society have been carried out as a prospective investigation on the prognosis of BS since 2001 in Japan. The annual incidence of cardiovascular events in patients with a history of VF and cardiac arrest was found to be similar in the reports from the USA and Europe, however, the prognosis of the patients’ group with a history of syncope was better than those in the western countries.

The indication of ICD implantation in patients with coved-type Brugada ECG without the induction of polymorphic ventricular tachycardia or VF by EPS is determined as class IIb even if there is a history of syncope. Regarding patients with no active application of ICD implantation, the family history, the presence of mutation of the myocardial Na channel (SCNSA) and the presence of late potential-positive findings are used as references in considering the applicability of ICD implantation, and a careful follow-up is carried out when implantation is not performed.

In recent years, ILR has been reported to be useful for the diagnosis of syncope, with a significantly higher diagnostic rate than other conventional tests. The Randomized Assessment of Syncope Trial (RAST) [10] was the first trial for idiopathic syncope with ILR. In this trial, 60 patients with idiopathic syncope and no abnormalities upon orthostatic blood pressure testing, Holter electrocardiography, and echocardiogram were randomly assigned to 30 cases of a conventional test group that underwent external loop recorder, tilt test, and electrophysiological testing, along with 30 cases of an ILR group. Each group was followed up for 12 months and cross-over between both groups when a diagnosis could not be made. The results demonstrated that the diagnosis was obtained with 52% for the ILR group in comparison with 20% for the conventional group. Following cross-over, ILR was more likely to result in an accurate diagnosis than conventional testing.

In this case, ILR revealed two episodes of VF both approximately 1 min in duration with spontaneous termination, and 2 mm elevation of the ST-segment with coved-type configuration was observed before an episode of VF which was not observed in baseline ECG. Baseline 12-lead ECG demonstrated the early repolarization in not only precordial leads but also inferolateral leads. Kamakura et al. [11] documented that Brugada-pattern ST elevation with inferolateral early repolarization was an indicator of poor prognosis and may represent electric heterogeneity in extensive regions of ventricles, which can result in ventricular arrhythmias. Furthermore, positive J wave deflection inscribed on the S wave.

![Fig. 4. Presentation of QRS morphology recorded by implanted loop recorder: (A) baseline electrocardiogram (ECG); (B and C) before first event (B) and second event (C); (D and E) after first event (D) and second event (E), ST-segment was elevated before first and second event as compared with baseline ECG. In addition, J-wave notch appeared just before episode of ventricular fibrillation (VF) (black arrows), which disappeared immediately after the termination of VF episode.](image-url)
emerged just before VF occurrence and disappeared after the termination of VF on the record of ILR in our case (Fig. 4). Sorgente et al. [12] reported the case of a patient with Brugada syndrome, in whom early repolarization in the inferolateral leads disappeared after a VF episode. They also demonstrated that 12-lead ECG obtained after VF termination revealed disappearance of J wave in inferolateral leads. These reports and our case may suggest the etiological association between the presence of early repolarization abnormalities and VF occurrence.

ICD implantation was performed in the present case on the basis of VF documentation by ILR. In this case, although there was a history of syncope, a family history of sudden death or the results of conventional test were negative, so the patient was initially classified as IIIb indication for ICD implantation. ILR was useful for detecting spontaneous episodes of lethal ventricular tachyarrhythmia. Our case suggests that ILCR implantation can provide additional diagnostic information in patients with BS whose applicability for ICD implantation is not conclusive by conventional tests.

References


