Journal of Arrhythmia 32 (2016) 70–73

Contents lists available at ScienceDirect

Journal of Arrhythmia

journal homepage: www.elsevier.com/locate/joa

Case Report

An overlap of Brugada syndrome and arrhythmogenic right ventricular cardiomyopathy/dysplasia

Shohei Kataoka, MD*, Naoki Serizawa, MD, Kazutaka Kitamura, MD, Atsushi Suzuki, MD, Tsuyoshi Suzuki, MD, Tsuyoshi Shiga, MD, Morio Shoda, MD, Nobuhisa Hagiwara, MD

Department of Cardiology, Tokyo Women's Medical University, 8-1 Kawadacho, Shinjuku-ku, Tokyo 162-8666, Japan

ARTICLE INFO

Article history:
Received 15 July 2015
Received in revised form
25 October 2015
Accepted 29 October 2015
Available online 30 November 2015

Keywords:
Brugada syndrome
Arrhythmogenic right ventricular cardiomyopathy/dysplasia
An overlap disease

ABSTRACT

Overlapping characteristics of Brugada syndrome (BrS) and arrhythmogenic right ventricular cardiomyopathy/dysplasia (ARVC/D) have been reported in recent studies, but little is known about the overlapping disease state of BrS and ARVC/D. A 36-year-old man, hospitalized at our institution for syncope, presented with this overlapping disease state. The electrocardiogram showed spontaneous coved-type ST-segment elevation, and ventricular fibrillation was induced by right ventricular outflow tract stimulation in an electrophysiological study. BrS was subsequently diagnosed; additionally, the presence of epsilon-like waves and right ventricular structural abnormalities met with the 2010 revised task force criteria for ARVC/D. After careful investigation for both BrS and ARVC/D, an implantable cardioverter defibrillator was inserted in the patient. This case revealed 2 important clinical findings: (1) BrS and ARVC/D clinical features can coexist in a single patient, and EPS might be useful for determining the phenotype of overlapping disease (e.g., BrS-like or ARVC/D-like). (2) An overlapping disease state of BrS and ARVC/D can change phenotypically during its clinical course. Therefore, careful examination and attentive follow-up are required for patients with BrS or ARVC/D.

1. Introduction

Brugada syndrome (BrS) is characterized by right precordial ST-segment elevation followed by a negative T wave and sudden cardiac death from ventricular fibrillation in patients with structurally normal hearts [1]. Recent studies have revealed structural or electrocardiographic abnormalities, such as right ventricular dilatation or epsilon-like waves, in some patients with BrS [2,3]. These abnormalities are commonly considered characteristics of arrhythmogenic right ventricular cardiomyopathy/dysplasia (ARVC/D). It is generally known that there are clinical similarities between BrS and ARVC/D [4]; however, little is known about the clinical features of patients with an overlapping disease state of BrS and ARVC/D, and accordingly, treatment of such patients remains poorly understood. In order to understand this condition better, we report here, a case of overlapping disease state of BrS and ARVC/D.

2. Case report

A 36-year-old man presented to our institution with syncope, and was subsequently, hospitalized. He had no family history of sudden cardiac death. The electrocardiogram (ECG) showed spontaneous coved-type ST-segment elevation (Brugada type 1 ECG); we initially suspected BrS. In an electrophysiological study (EPS) conducted prior to pilsicainide infusion, neither ventricular tachycardia (VT) nor ventricular fibrillation (VF) was induced by a single or double stimulus to the right ventricular apex or right ventricular outflow tract. A single stimulus to the right ventricular outflow tract during pilsicainide infusion induced ventricular fibrillation, which was thereafter, inhibited during isoproterenol infusion. Therefore, the patient was diagnosed with BrS, although the clinical presentation differed from typical BrS to some extent. First, the chest radiograph and cardiovascular magnetic resonance imaging (CMR) showed mild right ventricular (RV) dilatation (Fig.1A, B). Second, RV angiography demonstrated RV dilatation and akinesis in the inferior wall (Fig.1C, D), although coronary angiography did not show critical stenosis, and the provocation test failed to induce coronary spasm. Finally, epsilon-like waves were seen in spontaneous type 1 ECG (Fig.2). As a result, the patient not only met the diagnostic criteria for BrS but also met the

* Corresponding author. Tel.: +81 3 3353 8111; fax: +81 3 3353 6793.
E-mail address: shoheikataoka0818@gmail.com (S. Kataoka).

http://dx.doi.org/10.1016/j.joa.2015.10.007
1880-4276/

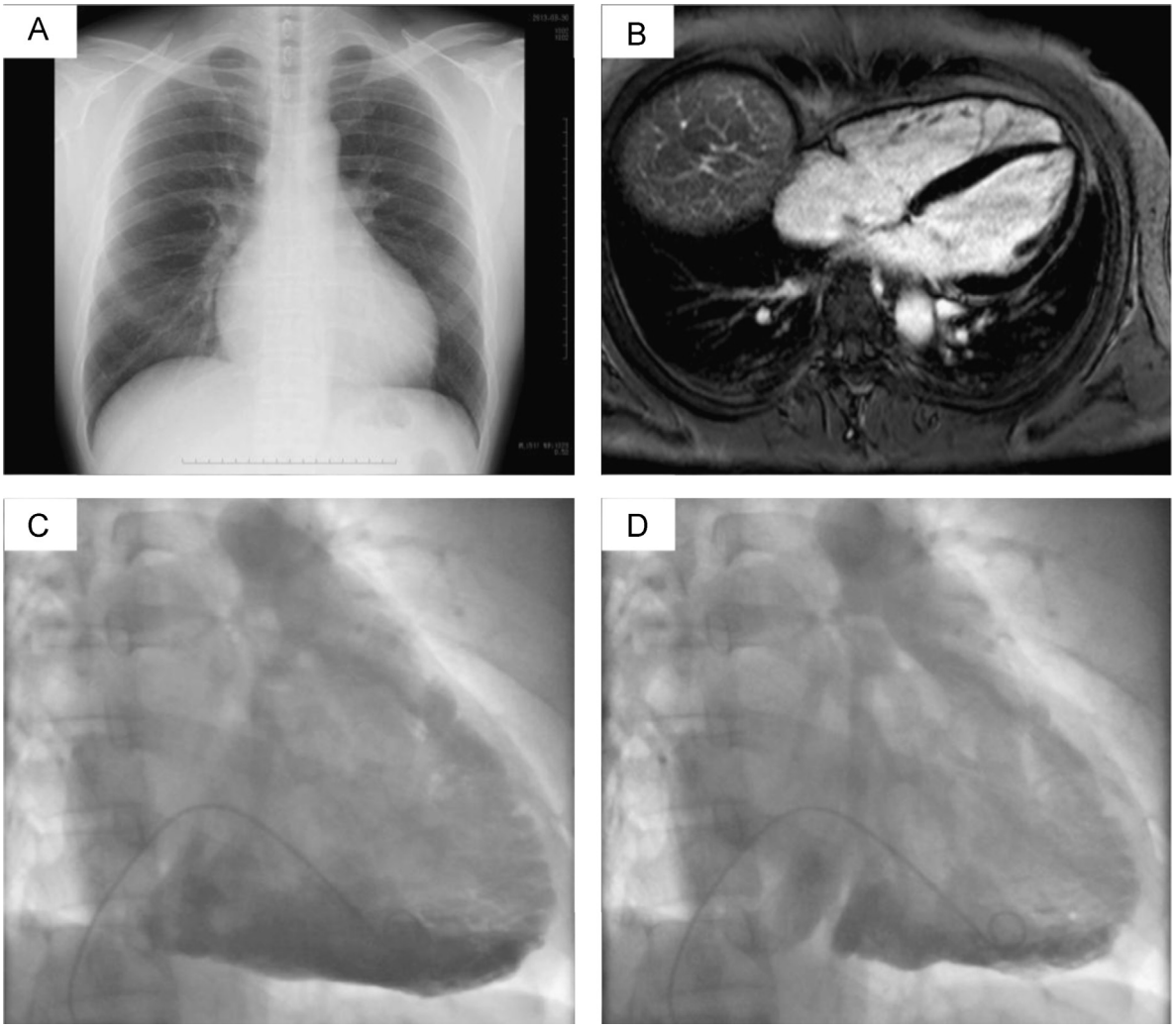

Fig. 1. (A)Chest radiograph reveals mild right ventricle dilatation. (B) Cardiovascular magnetic resonance imaging shows mild right ventricle dilatation and no late gadolinium enhancement. (C) Right ventricular angiography demonstrates right ventricular dilatation and akinesis in the inferior wall. (D) Computed tomography does not reveal fatty change in the right ventricular myocardium.

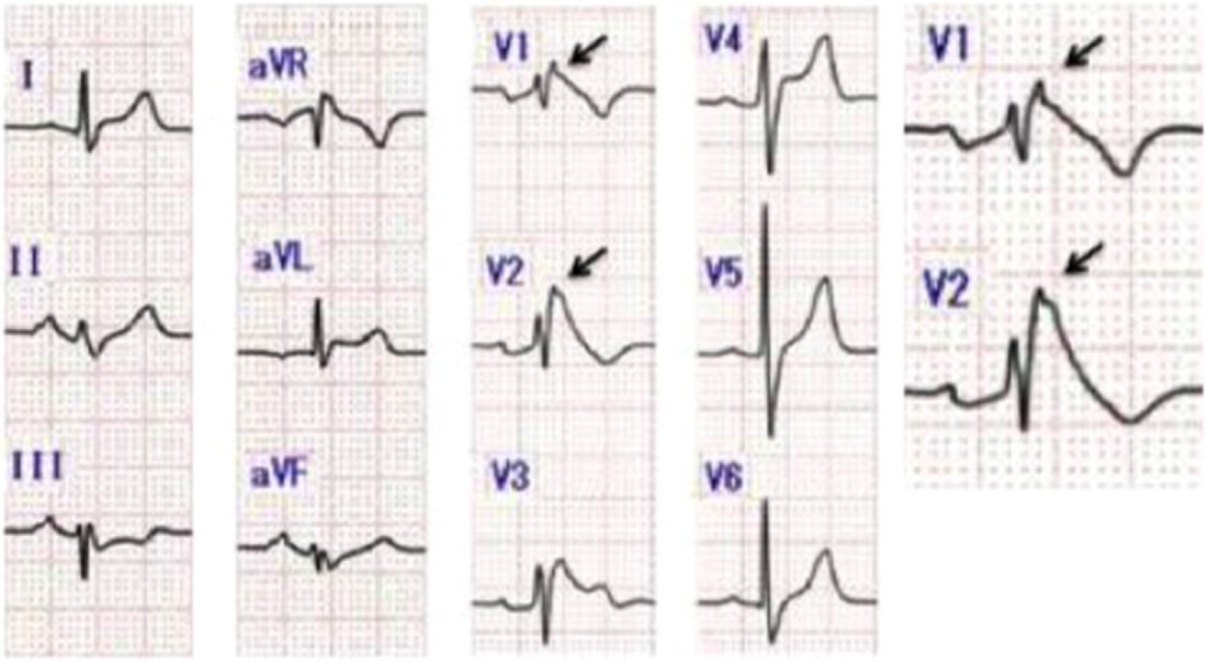

Fig. 2. The patient's electrocardiogram shows spontaneous coved-type ST-segment elevation and epsilon-like waves in type 1 ECG.