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Takotsubo Cardiomyopathy With Significant Coronary Stenosis and Atrioventricular Conduction Block: A Rare Case Report With 3 Year Follow-Up

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Introduction: Takotsubo cardiomyopathy (TCMP) is a rare acute cardiomyopathy characterized by acute chest pain syndrome, similar to myocardial infarction, except that no significant stenosis is observed on coronary angiography in patients with this condition; these findings aid the diagnosis of TCMP.

Case Presentation: We discuss an unusual case of TCMP in a 45-year-old woman with complete heart block and significant coronary artery stenosis. Maximal exercise test and perfusion scan after 1 month from the acute event did not show any ischemia; therefore, revascularization was not recommended. Her follow-up with normal echocardiographic data 3 years after the first event showed no recurrence

Conclusions: The present case and a few previous cases have showed that severe coronary artery disease may be occur in patients with TCMP and that TCMP may be associated with a high-degree atrioventricular block. The association between atrioventricular conduction block and TCMP as well as significant coronary stenosis is rarely reported; therefore, coronary angiography should be performed in all patients with clinical TCMP and the previous definition should be reconsidered. The occurrence of arrhythmia and later recovery is expected in these patients (due to a catecholamine surge).

Keywords: Takotsubo Cardiomyopathy; Complete Heart Block; Coronary Artery Disease

1. Introduction

Takotsubo cardiomyopathy (TCMP), also known as broken heart syndrome, stress cardiomyopathy, and transient apical ballooning syndrome, was previously defined as a rare cause of transient left ventricular systolic dysfunction with absence of significant coronary stenosis (1-3). On echocardiographic or left ventricular (LV) angiographic evaluation, dyskinesia or akinesia of apical segments and hyperkinesia of the base of the LV as a compensatory mechanism are observed. The term originated from Japanese, in which it means an octopus trap called "takotsubo"; however, reverse TCMP with hypokinesia or akinesia of basal and midventricle and normal apical segments was reported (1-3). Patients with this typical form of this cardiomyopathy present with a rise in the level of cardiac enzymes and changes in electrocardiographic (ECG) findings. TCMP is usually related to coronary vasospasm and microvascular disease secondary to high levels of catecholamine release; it is most commonly observed in middle-aged women and is completely reversible with supportive treatments (4). Complete heart block (CHB) with TCMP has been rarely reported in previous studies (5, 6).

We report a rare case of TCMP with significant coronary stenosis (70% stenosis in the left anterior descending artery, Figure 1) with transient CHB in a 45-year-old woman with a history of neurosis and controlled hypertension, as well as findings through a 3-year follow-up.

2. Case Presentation

We present the case of a 45-year-old Iranian woman with acute dyspnea and atypical chest pain who was referred to our emergency department. She had a past history of neurosis and relatively controlled hypertension. Her vital signs were stable on admission, except for blood pressure, which was 160/90 mmHg. A standard ECG showed diffuse, new ST-segment elevation (about 3 mm) in the anterolateral (V4-V6) and the inferior leads (II, III, and aVF) with normal sinus rhythm and transient arrhythmias (atrioventricular dissociation, infrequent premature ventricular contractions [PVCs], and complete heart block) (Figure 2).

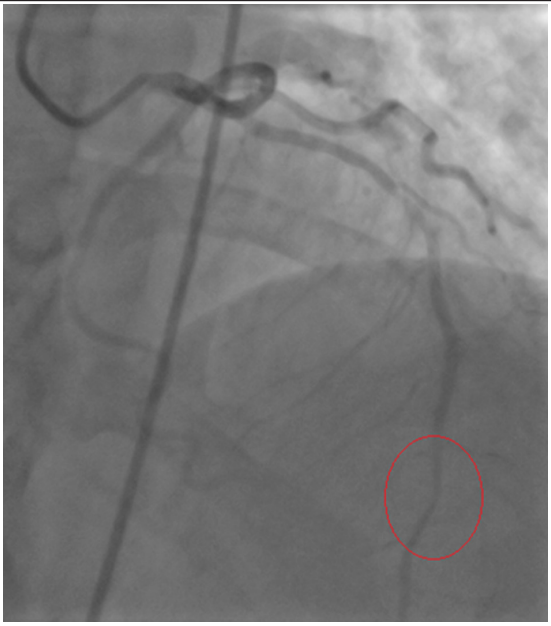

Figure 1. Coronary Angiography With Two Significant Stenosis at the Proximal and the Middle Parts of Left Anterior Descending Artery

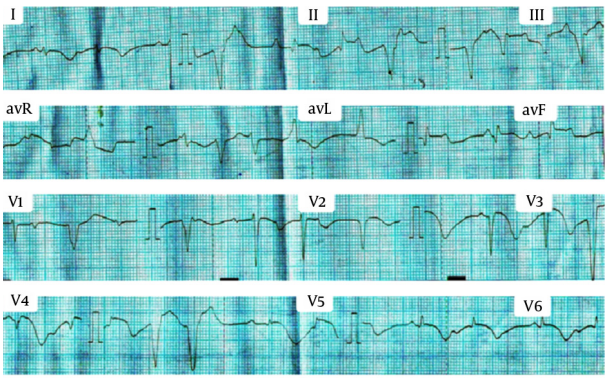

Figure 2. Electrocardiogram in the Acute Phase Showing Diffuse ST-Segment Elevation in the Anterolateral and the Inferior Leads and Normal Sinus Rhythm With Severe Arrhythmia (Atrioventricular Dissociation, Infrequent PVC, and Complete Heart Block)

Findings on laboratory tests were within the normal range, except for marked elevation in the level of cardiac enzymes (creatine kinase [CK] MB and troponin).

The level of CK MB was about 151 UI/L (normal level: < 25UI/L) and the level of troponin T was about 3 ng/mL (normal level: < 0.02 ng/mL). Emergent coronary angiography was performed, and significant lesions wrapped around the left anterior descending artery at the proximal and the middle parts were observed. Left ventriculography showed left ventricle systolic dysfunction (left ventricular ejection fraction [LVEF] of about 30% - 35%) and apical ballooning pattern (severe akinesia in apical segments and normal motion in basal segments), and mild mitral regurgitation (MR) (Figure 3).

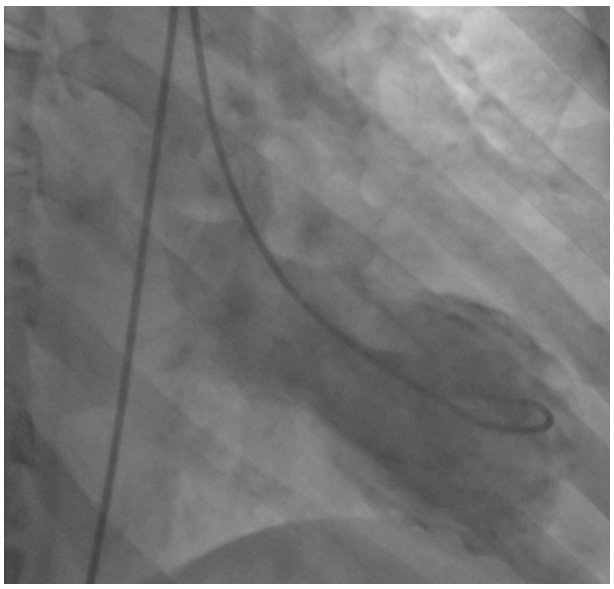

Figure 3. Left Ventriculography Showing Apical Ballooning With Left Ventricle Systolic Dysfunction

Echocardiography also showed the same pattern that was consistent with findings for TCMP. Temporary pacemaker was not inserted as there was recovery from CHB and AV dissociation after 5 days of admission, and the patient was stable during admission.

The patient was treated with aspirin, angiotensin-converting enzyme inhibitor, diuretic, clopidogrel, and nitrates, but beta blockers were not administered due to presence of CHB. The patient's hospital course was uncomplicated, and she was reevaluated after 7 days. Echocardiographic data showed improvement of apical wall motion abnormality, and LV ejection fraction had increased to 45% and ECG showed normal sinus rhythm with a heart rate of about 73 beats/min with no arrhythmia; the patient was discharged from the hospital.

Repeated echocardiography after 3 weeks showed normal LV systolic function; the LVEF was about 55% with no wall motion abnormality. ECG showed normal sinus rhythm and a narrow complex QRS. Her heart rate was about 75 - 80 beats/min with asymptomatic Wenckebach block. Maximal exercise stress test and perfusion scan performed after 1 month from the acute event did not show any ischemia; hence, revascularization was not recommended. During the 3 years of follow-up (2012 - 2014), no events occurred. However, after the second year, a Wenckebach block occurred, with an acceptable heart rate of 72 beats/min on ECG, and echocardiography data were normal. At the third year of follow-up, a first-degree atrioventricular block was noted with a heart rate of about 70 beats/min on ECG (Figure 4) and echocardiography findings were normal (LVEF=55%); no complaint or symptom was reported during this time, and the patient was treated with diuretic and enalapril (for hypertension), aspirin, atorvastatin.

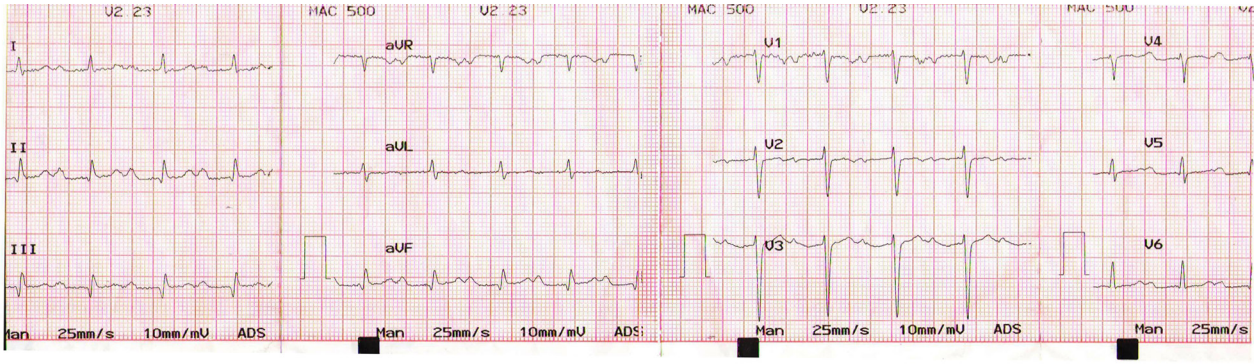

Figure 4. Electrocardiogram Showing Normal Sinus Rhythm With First-Degree Atrioventricular Block Recorded 3 Years Later

3. Discussion

TCMP is a rare, acute, reversible cardiomyopathy that is characterized by acute chest pain syndrome similar to myocardial infarction, and shows no significant coronary stenosis on angiography; the diagnosis of this finding was previously considered challenging (1-3). The exact mechanism underlying this condition is not known. It may be related to neurocardiogenic myocardial disease and excess stimulation of beta-1 sympathetic receptors in the setting of acute severe physiologic or emotional stress (4) and coronary vasospasm, plaque rupture, and thrombus formation with spontaneous recanalization (7). Patients are predominantly postmenopausal women, but TCMP is also reported premenopausal patients such as in the present patient.

Most previous studies have shown that the absence of epicardial coronary obstruction and obstructive CAD is a rule in TCMP diagnosis and the presence of CAD in TCMP was considered an incidental finding in high-risk patients (8)." Some studies have reported that TCMP is not a distinct clinical entity, but rather a manifestation of aborted anterior myocardial infarction in patients with a long wraparound left anterior descending artery transient occlusion in the left anterior descending artery and subsequent spontaneous thrombus lysis and apical stunning and wall motion abnormalities that show improvement during follow-up after treatment (8). A review of the literature showed that although ischemia is transient in TCMP patients, an atrioventricular conduction defect with late recovery is observed (5, 6). A rare case of persistent complete heart block that needed a permanent pacemaker was also reported (mostly due to a concealed disease in the atrioventricular conduction system before the onset of this cardiomyopathy) (5, 6); however, in this case, a transient high-degree atrioventricular block and a change in AV conduction (from the third to the second and the second to the first AV block) during the 3-year follow-up confirmed the functional atrioventricular block due to vagal storm in the present case.

We present an unusual case of TCMP with a coexisting severe LAD lesion, presenting with acute myocardial infarction-like, third-, second-, and first-degree atrioventricular block during 3 years of follow-up. As there was no ischemia observed on a perfusion scan performed 1 month after the event, spontaneous thrombus lysis or spasm was considered in this patient, similar to that reported in previous studies. A neurocardiogenic mechanism was suspected in this patient, as there was a functional, transient atrioventricular block.

The association between atrioventricular conduction block and TCMP as well as a significant coronary stenosis has rarely been reported.

Therefore, evaluation for concomitant coronary artery disease and follow-up for arrhythmia in clinically indicated TCMP is suggested, and late recovery of atrioventricular conduction abnormality is expected and further investigation on TCMP is needed.

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Authors' Contributions

Hakimeh Saadatifar managed the patient and helped in writing the first draft of the manuscript. Samira Saadatifar and Fahimeh Khoshhal Dehdar wrote the entire manuscript, and Maryam Moshkani Farahani rewrote and revised the manuscript.

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