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Case Report

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Recurrent midventricular ballooning, a rare phenomenon

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Introduction

ABSTRACT

Takotsubo cardiomyopathy is characterized by transient systolic ballooning of apical or mid segments of the left ventricle (LV) mimicking acute myocardial infarction in the absence of obstructive epicardial coronary artery disease. Recurrences remain rare and most recurrence case reports are about apical ballooning or typical Takotsubo cardiomyopathy. We present a rare case of recurrence in mid ventricular ballooning or atypical variant of Takotsubo cardiomyopathy. This case highlights the importance of future follow-up and potential of recurrence even in atypical variants.

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Takotsubo cardiomyopathy (TCM), also known as stress cardiomyopathy was first recognized as a separate clinical entity in Japan [1]. In the past decade its incidence has been established in western populations as well [2–5]. TCM is characterized by transient systolic ballooning of apical or mid segments of the left ventricle (LV) mimicking acute myocardial infarction in the absence of obstructive epicardial coronary artery disease.

Although it is generally considered a benign phenomenon and recurrence remains rare (0-8%) [6–9], it is important to recognize its potential complications and recurrence which could have serious consequences including death [10].

Most recurrence case reports are about apical ballooning or typical TCM. We present a rare case report of recurrence in mid ventricular ballooning or atypical variant of TCM. This case highlights the importance of future follow-up and potential of recurrence even in atypical TCM.

Case report

A 53-year-old female nursing director presented with new onset of typical angina pectoris at rest associated with palpitations. The symptoms lasted intermittently for 5 h prior to the presentation. The patient had no history of similar symptoms in the past; there was no syncope, dyspnea, or neurological or gastroenterological symptoms. She reported significant job stress. Past medical history was significant for dyslipidemia, hypertension, and hypothyroidism. Medications prior to admission included aspirin, ramipril, levothyroxine, hydrochlorothiazide, and fish oil. There was no history of smoking, alcohol, or drug abuse.

The initial physical exam was unremarkable except blood pressure of 140/90 mmHg.

Electrocardiography (ECG) showed sinus rhythm and ST-T changes in precordial leads (Fig. 1). The chest pain resolved after treatment with nitrates, beta blocker, and heparin.

Urgent angiogram demonstrated minor, nonobstructive coronary disease (30% mid left anterior descending artery lesion) (Figs. 2 and 3). Left ventriculogram showed akinesis to dyskinesis of mid left ventricular segments with hyperkinetic basal and apical segments (Fig. 4a and b). Left ventricular ejection fraction (LVEF) was 40%. Transthoracic two dimensional echocardiogram (ECHO) confirmed the wall motion abnormalities and showed no significant valvular heart disease. Initial troponin was minimally elevated with peak troponin 4.09 ng/mL (0.00–0.01 ng/mL is normal range). ST segment changes resolved on follow up ECG after one day.

She had occasional chest pains and palpitations but no new ECG changes post cardiac catheterization. She was discharged on angiotensin-converting enzyme inhibitor, beta blocker, statin, nitrates, and aspirin. Repeat ECHO one month later showed complete resolution of LV wall motion abnormalities and LVEF of 65%.

Five months later the patient experienced recurrent angina pectoris for over an hour at rest and was noted to have similar to prior presentation ECG changes. The initial troponin was 0.33 ng/mL and it peaked at 5.26 ng/mL. She was treated with nitrates, enoxaparin, and continued on beta blocker and aspirin. ECHO showed mid ventricular regional wall motion abnormalities with hypercontractile apex and base segments and LVEF of 50% (Fig. 5a and b). Coronary

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Fig. 1. Electrocardiogram at presentation.



Fig. 2. Normal left coronary angiogram.

angiography showed no change in coronary anatomy compared to the previous study. The same medications were continued, and the patient was counseled about stress management. She has remained mostly asymptomatic except for occasional chest discomfort for the past 2 years of follow up.

Discussion

TCM is generally considered reversible and LV function usually recovers within one month but several complications such as cardiogenic shock, ventricular arrhythmias, left ventricular rupture, and death have also been reported in the past [10].

Recurrence of TCM is relatively rare. Most case reports of recurrences are of typical TCM; it remains extremely rare in atypical variants [5,6,11]. Due to limited data in the literature on TCM follow up (variable lengths from a few days to 4 years) the exact incidence of recurrence is still not well established [7–9].

Most patients are prescribed cardioprotective drugs recommended for acute coronary syndrome but relapses have occurred and no data exist about efficacy of these drugs in TCM [5,7,12]. There are no established predictors of recurrence, although in most patients internal or external stressors seem to be the triggers [2,5,12,13]. Therefore, possible improvement in patient's psychosocial dynamics might prevent the recurrences. Professional psychiatry evaluation, pharmacologic, and non-pharmacologic stress management could play a positive role in such patients.

The understanding of pathophysiology, etiology, treatment, clinical predictors, and prevention is in development and many questions remain unanswered. Several mechanisms have been proposed. These include myocardial stunning secondary to



Fig. 3. Right coronary angiogram.



Fig. 4. Left ventriculogram first presentation. (a) Systole first presentation (with border tracing). (b) Diastole first presentation.

catecholamine storm, multivessel coronary spasm, abnormal microvascular function [6,10,13], ruptured plaque, and genetic predisposition [14]. Mostly internal and external stressors trigger it, with more predilections in postmenopausal women [5].

Our patient has had a typical clinical presentation of the stress cardiomyopathy both on initial and recurrent presentation. She belonged to the typical age group and gender (post-menopausal women) and admitted to continued and severe emotional stress that has been known to trigger this syndrome. But atypical TCM features on ECHO and left ventriculogram and its recurrence in our patient may present a unique opportunity to improve our understanding of this phenomenon.

The theory of diffuse spasm appears less likely as an etiology in case of mid left ventricular ballooning. One would have to postulate a very specific, yet multi vessel spasm at "same level" of longitudinal axis of the left ventricle, but then sparing of the apex would be difficult to explain. Coronary spasm causing the mid ventricular motion abnormality would be expected to cause even more pronounced abnormality distally, in the apex, which was not the case in our patient. Additionally, it has been shown previously that perfusion mismatch seen in TCM does not correlate with single epicardial vessel [2]. This has led some to believe that perhaps impaired coronary microvascular function is the possible cause of TCM [6].

It has been proposed that certain anatomical features of LV, such as sigmoid interventricular septum, small left ventricle outflow tract (LVOT), and abnormally oriented sagging mitral valve apparatus might cause LVOT obstruction in settings of intense adrenergic stimulation or hypovolemia [15,16]. Due to transient LVOT obstruction increased pressure and stress on anterior wall cause increased oxygen consumption and result in perfusion mismatch and ballooning. We speculate further that since many patients with TCM in previous studies including ours had hypertension [5,6] that might change cardiac myocyte physiology and contribute to change in LV function causing LVOT obstruction. However, in our case we have not discovered any significant gradients across LVOT.

Direct toxic effect of catecholamines on myocardium has been postulated as etiology of stress cardiomyopathy [13] with evidence of higher adrenergic receptors density in the LV apex in cases of apical ballooning. Variation in anatomic distribution of beta adrenergic receptors in LV segments and difference in their sensitivities to catecholamines could lead to variation in response of LV regions. It would be interesting to study the receptor density/sensitivity in the variants of TCM, especially in recurrent cases. However, there are case reports where different LV segments were involved in recurrent TCM episodes [17,18]. Then, unless receptor density and their sensitivity change with time, this hypothesis appears difficult to explain. It is also possible that mid ventricular and apical ballooning syndrome could have different and more than one etiology.

In summary, our case represents a rare clinical entity of recurrent atypical TCM with only few such cases reported before. Increased awareness among the medical community has improved



Fig. 5. Two-dimensional echocardiogram second presentation. (a) Systole second presentation. (b) Diastole second presentation.

our ability to identify more cases in the past decade. This case however brings forward a unique and so far less recognized fact that TCM in its atypical form can also be recurrent.

We recommend a longer follow up in all these patients to improve our understanding of disease process. We also believe that stress prevention and management could have a role in control of future relapses.

Conflict of interest

All authors have no conflicts of interest that should be disclosed.

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