Cor et Vasa

Available online at www.sciencedirect.com

SciVerse ScienceDirect

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

Kasuistika | Case report

Biventricular Takotsubo syndrome in a patient with coronary abnormality and end-stage renal disease

Ferit Onur Mutluer[a], Rosalinda Madonna[b,c], Cihan Cevik[b], Maher M. Nasser[b], Raffaele De Caterina[c]

[a] *Department of Cardiology, Siyami Ersek Cardiovascular Surgery Center, Istanbul, Turkey*
[b] *Texas Heart Institute and St. Luke's Episcopal Hospital, Houston, TX, USA*
[c] *Institute of Cardiology, G. d'Annunzio University, Chieti, Italy*

ARTICLE INFO

Article history:
Received: 5 September 2012
Accepted: 13 September 2012
Available online:
28 September 2012

Keywords:
Apical ballooning
Coronary artery anomalies
End-stage renal disease
Takotsubo syndrome

ABSTRACT

The Takotsubo syndrome is a clinical entity consisting of transient, regional left ventricular (LV) contraction abnormalities in the absence of significant epicardial coronary artery disease. Patients usually present with acute hemodynamic deterioration following an emotional or physical stress. The Takotsubo syndrome is uncommon among patients with end-stage renal disease (ESRD) and patients with congenital coronary abnormalities, such as right coronary artery (RCA) originating from left sinus of Valsalva. Here we describe a patient presenting with acute respiratory distress and anterolateral ST–T segment changes, with negative troponin-I and elevated levels of brain natriuretic peptide. Coronary angiography showed a right coronary artery arising from the left sinus of Valsalva without obstructive coronary artery disease, while ventriculography and echocardiogram showed findings compatible with apical ballooning. Magnetic resonance imaging confirmed the diagnosis of a biventricular Takotsubo cardiomyopathy.

Klíčová slova:
Anomálie koronárních tepen
Stresová kardiomyopatie
Tako-tsubo kardiomyopatie
Terminální selhání ledvin

SOUHRN

Tako-tsubo (stresová) kardiomyopatie je klinickou jednotkou, při níž dochází k přechodným, regionálním abnormalitám stahů levé komory (LK) v nepřítomnosti významného postižení epikardiálních koronárních tepen. K lékaři se pacienti obvykle dostavují s akutním hemodynamickým zhoršením po emocionální nebo fyzické zátěži. U nemocných s terminálním selháním ledvin a u osob s vrozenými anomáliemi koronárních tepen, např. s pravou věnčitou tepnou odstupující z levého Valsalvova sinu, se tako-tsubo kardiomyopatie běžně nevyskytuje. V našem příspěvku popisujeme pacienta s akutní dechovou tísní a změnami anterolaterálního segmentu ST–T, negativním troponinem I a zvýšenými hodnotami natriuretického peptidu typu B. Koronarografie zjistila anomální odstup pravé věnčité tepny z levého Valsalvova sinu bez přítomnosti obstrukční ischemické choroby srdeční, zatímco nálezy ventrikulografického a echokardiografického vyšetření ukazovaly na stresovou kardiomyopatii. Vyšetření magnetickou rezonancí potvrdilo diagnózu biventrikulární tako-tsubo kardiomyopatie.

Address: Raffaele De Caterina, MD, PhD, FESC, Institute of Cardiology, G. d'Annunzio University, Chieti, C/o Ospedale SS. Annunziata, Via dei Vestini, 66013 Chieti, Italy, e-mail: rdecater@unich.it
DOI: 10.1016/j.crvasa.2012.09.005

Please cite this article as: F.O. Mutluer, et al., Biventricular Takotsubo syndrome in a patient with coronary abnormality and end-stage renal disease, Cor et Vasa 55 (2013) e277–e280 as published in the online version of *Cor et Vasa* available at http://www.sciencedirect.com/science/article/pii/S0010865012001099

The Takotsubo syndrome is a clinical entity consisting of transient, regional left ventricular (LV) contraction abnormalities in the absence of significant epicardial coronary artery disease. Classical presentation consists of an acute hemodynamic deterioration following an emotional or physical stress, typically in a postmenopausal woman. Critical illness, hypertension, chronic obstructive pulmonary disease and bronchial asthma [1] appear to increase the risk of Takotsubo syndrome. Such diagnosis seems to be uncommon among end-stage renal disease (ESRD) patients, and has not been reported in patients with congenital coronary artery abnormalities. The absence of stress as a trigger is also uncommon. In this report we describe for the first time the occurrence of a biventricular Takotsubo syndrome in a patient with ESRD, subclinical hypothyroidism, and a right coronary artery (RCA) arising from the left sinus of Valsalva in the absence of a history of physical or emotional stress. The atypical features of this case make it a rather unique presentation for the Takotsubo syndrome never previously described.

A 69-year-old African-American woman with known history of hypertension, diabetes mellitus and ESRD due to hypertensive nephrosclerosis and currently on a hemodialysis program was admitted to the emergency room (ER) with acute respiratory distress. The patient was hypertensive (200/90 mmHg), tachycardic (120 b.p.m.), with a respiratory rate of 30/min and a normal body temperature (36.6 °C). The physical examination was unremarkable, with the exception of bilaterally decreased breath sounds and a third heart sound (S3). Arterial oxygen saturation was 84%. The ECG showed anterolateral ST–T segment changes (Fig. 1, panel A1). The patient was intubated and admitted to the Coronary Care Unit. Admission cardiac troponin-I value was < 0.15 ng/mL. Troponin value peaked at 3.45 ng/mL, and then decreased. At the same time, brain natriuretic peptide (BNP) level was 2356 pg/mL. A routine set of biohumoral examinations yielded normal results, with the exception of a creatinine level of 4.63 mg/dL and a blood urea nitrogen (BUN) level of 29 mg/dL. Because of the ECG changes suggestive of an anterolateral ST elevation myocardial infarction (STEMI), the patient was transferred to the cath-lab. Coronary angiography showed a normal left coronary artery (Fig. 1, panel B1) and a right coronary artery arising from the left coronary sinus (Fig. 1, panel B2), while ventriculography showed findings compatible with apical ballooning (Fig. 1, panel C).

Transthoracic echocardiography showed mild global left ventricular dysfunction (left ventricle ejection fraction, LVEF = 45–49%), a dyskinesia in the mid to distal segments of both ventricles, a suspicious trabeculation in the apex and the apical segments of the left ventricle in the apical 4-chamber view, and a mild pericardial effusion localized close to the posterobasal wall (Fig. 1, panels D1 and D2).

A cardiac magnetic resonance imaging (MRI) study was prescribed to rule out LV noncompaction cardiomyopathy. The MRI confirmed the diagnosis of a biventricular Takotsubo cardiomyopathy and ruled out LV noncompaction. The distal half of both the left and the right ventricles were found to be dyskinetic (Fig. 2, panels A1 and A2). T2-map sequences were suggestive of apical edema (Fig. 2, panel B).

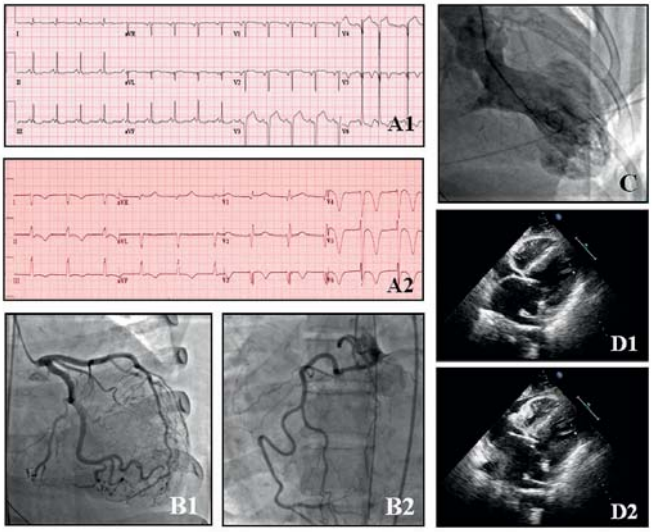

Fig. 1 – Admission ECG (panel A1), showing anterolateral ST–T segment changes, which prompted coronary angiography; panel A2, ECG at follow-up; panel B, left (panel B1) and right (panel B2) coronary angiography, showing the right coronary artery arising from the left coronary sinus; (panel C) ventriculography, demonstrating a typical apical ballooning; (panel D) subcostal 4-chamber echocardiographic views at end-diastole (panel D1) and end-systole (panel D2), confirming the presence of biventricular involvement.

In the follow-up, the patient developed fatigue. No dyspnea, orthopnea, chest pain or palpitations were reported. After 7 days from admission there was no change in the ECG (Fig. 1, panel A2). A chest X-ray demonstrated mild pleural effusion. In a control echo at the same time, there was a mildly increased anteroseptal, apicoseptal and apical hypokinesis, and a mild mitral regurgitation, while pericardial effusion had disappeared.

The usual presentation of the Takotsubo syndrome is an acute-onset LV dysfunction following a physical or emotional stress in a female postmenopausal patient.

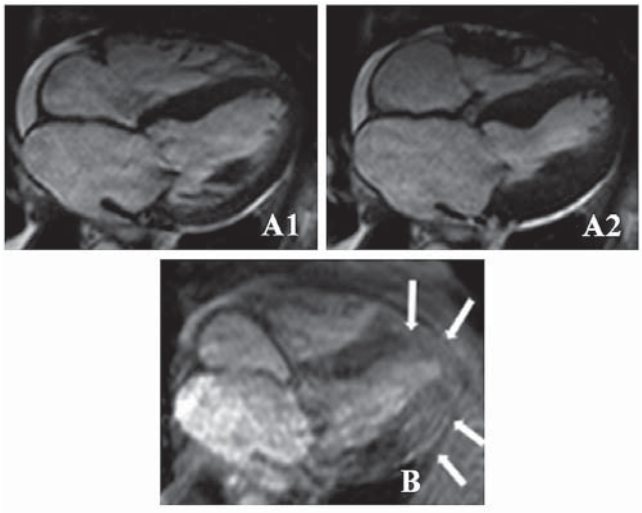

Fig. 2 – Magnetic resonance imaging (MRI), T2-map sequences; panel A, 4-chamber end-diastolic (panel A1) and end-systolic images (panel A2); (panel B) apical edema (arrows).

The most commonly accepted mechanism is endogenous catecholamine toxicity. A recent emotional or physical stress, histologic changes in the myocardium similar to those of catecholamine toxicity in animal studies, and catecholamine levels significantly higher than in patients with MI all support this hypothesis. A transient coronary vasospasm leading to myocardial stunning is incompatible with the usual unresponsiveness of coronary arteries to intracoronary vasoconstrictors such as ergometrine or acetylcholine, and the rarity of a documentation of coronary vasospasm in patients undergoing cardiac catheterization in the acute phase supports the low likelihood of vasospasm as a pathogenetic mechanism. Another hypothesis is of a microcirculatory abnormality, which is supported by studies demonstrating significantly higher Thrombolysis In Myocardial Infarction (TIMI) counts in all coronary arteries [1].

There are few descriptions of the Takotsubo syndrome in patients with ESRD [2–6]. In each of these cases, a physical or emotional stress preceded the onset. ESRD patients might be, however, at increased risk of developing the Takotsubo cardiomyopathy because of increased sympathetic nerve activity in patients with ESRD independent of uremic toxins or volume disturbances [7]. Uremia also independently increases sympathetic outflow in patients with ESRD. The absence of any previous physical or emotional stress in the medical history also supports this hypothesis in our patient.

Biventricular Takotsubo syndrome is being reported more commonly over time. In a retrospective study, 8 out of 25 patients had a right ventricular involvement, and this was associated with significantly higher incidences of severe heart failure and intubations [8].

A right coronary artery originating from the left coronary sinus is a rare condition with an incidence of 0.05–0.1% [9]. Our patient was found to have an anomalous RCA originating from a separate ostium in the left coronary sinus. The ECG findings and the occurrence of areas of regional wall motion abnormality on transthoracic echocardiography are inconsistent with perfusion abnormality in myocardial areas supplied by the RCA. In addition, there is no described association between the Takotsubo syndrome and an anomalous origin of the RCA, ESRD and hypothyroidism. The possibility that all these conditions exist independently in a patient is lower than the possibility of some interplay of molecular and genetic mechanisms associated with this conditions and causing the clinical picture observed. Further research is therefore needed on this topic.

Disclosure statement

The authors report no financial relationships or conflicts of interest regarding the content of this manuscript.

References

[1] A.K. Sharma, J.P. Singh, E.K. Heist, Stress cardiomyopathy: diagnosis, pathophysiology, management, and prognosis, Critical Pathways in Cardiology 10 (2011) 142–147.
[2] T. Kusaba, H. Sasaki, T. Sakurada, et al., Takotsubo cardiomyopathy thought to be induced by MRSA meningitis and cervical epidural abscess in a maintenance-hemodialysis patient: case report, Nihon Jinzo Gakkai shi 46 (2004) 371–376.
[3] M. Fukui, Y. Mori, S. Tsujimoto, et al., 'Takotsubo' cardiomyopathy in a maintenance hemodialysis patient, Therapeutic Apheresis and Dialysis 10 (2006) 94–100.
[4] F. Takemoto, N. Chihara, N. Sawa, et al., Takotsubo cardiomyopathy in a patient undergoing hemodialysis, Kidney International 76 (2009) 467.
[5] Y.-T. Tsao, C.-S. Lin, S.-H. Lin, Thyrotoxicosis factitia induces Takotsubo cardiomyopathy in end-stage renal disease: a pathogenetic hypothesis, Kidney International 77 (2010) 468.
[6] S. Hassan, F. Hassan, D. Hassan, et al., Takotsubo cardiomyopathy associated with peritonitis in peritoneal dialysis patient, Renal Failure 33 (2011) 904–907.
[7] M. Hausberg, M. Kosch, P. Harmelink, et al., Sympathetic nerve activity in end-stage renal disease, Circulation 106 (2002) 1974–1979.
[8] D. Haghi, A. Athanasiadis, T. Papavassiliu, et al., Right ventricular involvement in Takotsubo cardiomyopathy, European Heart Journal 27 (2006) 2433–2439.
[9] A.C. Leme Neto, R.G. Carvalho, R.J. Rauen Jr, et al., Anomalous origin of the right coronary artery: diagnosis and treatment, Arquivos Brasileiros de Cardiologia 90 (2008) e9–e12.