

# Cuban Society of Cardiology Case Report



# Postoperative Takotsubo syndrome with severe left ventricular dysfunction following mitral valve replacement

Gustavo de J. Bermúdez Yera<sup>1</sup>, MD, MSc; Ernesto Chaljub Bravo<sup>1</sup>, MD; Yoandy López de la Cruz<sup>1</sup>, MD, MSc; Luis F. Vega Fleites<sup>2</sup>, MD; Alay Viñales Torres<sup>3</sup>, MD; Leonel Fuentes Herrera<sup>4</sup>, MD, MSc; and José R. Pérez de la Paz<sup>5</sup>, MD

Este artículo también está disponible en español

### ARTICLE INFORMATION

Received: April 28, 2019 Accepted: May 23, 2019

#### Competing interests

The authors declare no competing interests

### **ABSTRACT**

Takotsubo syndrome was first described in Japan in late 1989 and early 1990 and featured a group of patients with transient apical dyskinesia and preservation of basal left-ventricular contractility, which resembled the vessel used by fishermen to catch octopus. It is usually seen in situations involving catecholamine release and its clinical picture mimics that of acute myocardial infarction. The case presented occurs, surprisingly, in the postoperative period of cardiovascular surgery and is probably the first one found in this circumstance in Cuba: perioperative ischemia after mitral valve replacement, which recovered within nearly 72 hours, although complete recovery occurred later. Angiographic images are shown.

Keywords: Takotsubo Cardiomyopathy, Cardiac surgery, Mitral valve replacement, Postoperative period

# Síndrome de Takotsubo con disfunción grave de ventrículo izquierdo en el postoperatorio de reemplazo valvular mitral

# **RESUMEN**

El síndrome de Takotsubo, descrito por vez primera en Japón a fines de 1989 y principios de 1990, caracteriza un conjunto de pacientes con discinesia apical transitoria y conservación de la contractilidad hacia la base del ventrículo izquierdo, que recuerda la forma de la vasija usada por los pescadores para atrapar pulpos. Habitualmente se observa en situaciones que impliquen elevación de catecolaminas y su cuadro clínico es similar al de un infarto agudo de miocardio. El caso que se presenta ocurrió, sorprendentemente, en el postoperatorio de una cirugía cardiovascular y probablemente sea el primero encontrado en Cuba en esta circunstancia: isquemia perioperatoria tras reemplazo valvular mitral, que se recuperó en aproximadamente 72 horas, aunque su recuperación total fue más tardía. Se muestran las imágenes angiográficas.

*Palabras clave:* Miocardiopatía de Takotsubo, Cirugía cardíaca, Reemplazo valvular mitral, Período postoperatorio

☑ GJ Bermúdez Yera Calle Cuba 610, e/ Barcelona y Capitán Velasco. Santa Clara, CP 50200. Villa Clara, Cuba. E-mail address: gustavodejesus@infomed.sld.cu

### INTRODUCTION

Also known as the broken heart syndrome, of apical dyskinesia and stress-

<sup>&</sup>lt;sup>1</sup>Department of Cardiovascular Surgery, <sup>2</sup>Department of Interventional Cardiology, <sup>3</sup>Department of Anesthesiology, and

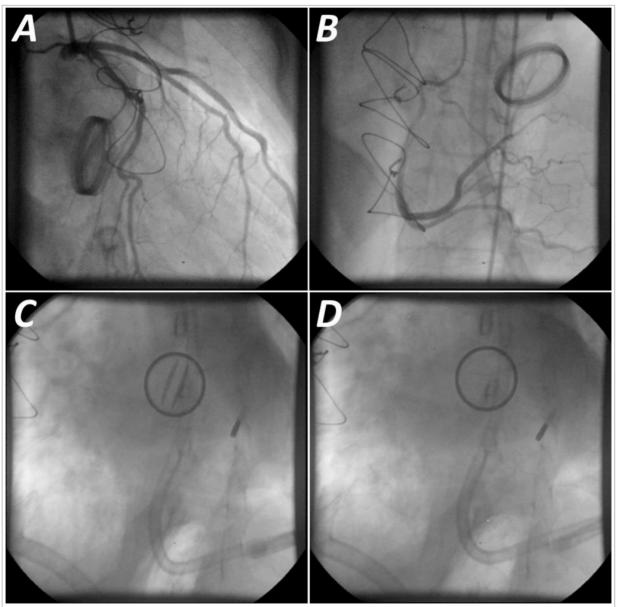
<sup>&</sup>lt;sup>4</sup>Intensive Care Unit, Cardiocentro Ernesto Guevara. Santa Clara, Villa Clara, Cuba.

<sup>&</sup>lt;sup>5</sup> Policlínico Universitario Santa Clara, Santa Clara, Villa Clara, Cuba.

induced cardiomyopathy, the Takotsubo syndrome was described for the first time in Japan, in 1990¹. It is characterized by transient systolic dysfunction of apical and middle segments of the left ventricle, with preserved contraction of its base, an aspect that –in the left ventriculography– mimics a container/trap traditionally used by Japanese for catching octopus, that is called Takotsubo.

The cardiomyopathy of the same name, most often affects postmenopausal women and routinely is caused by an intense emotional stress<sup>1</sup>. The American Heart Association (AHA) recognized the Takotsubo syndrome, in 2006, as a type of acquired cardiomyopathy<sup>2</sup>.

Its etiology and clinical features are not yet fully clarified<sup>3</sup>. Many trigger factors have been identified: psychological, exacerbation of systemic disease, neurogenic, pulmonary, gastrointestinal, renal disease and other nonspecific factors. Despite its unknown etiology, exaggerated sympathetic stimula-



**Fig. 1.** Angiographic and fluoroscopic images showing the normal left (**A**) and right (**B**) coronary arteries, the normofunctional mitral prosthetic valve —open in diastole (**C**) and closed in systole (**D**)—, and the unscathed sternal suture.

tion has been proposed as the central factor of its pathophysiology<sup>3,4</sup>. Patients with this syndrome have higher levels of catecholamine than patients with myocardial infarction with the same Killip class<sup>4</sup>.

The incidence in Latin America was unknown since it was initially described in the Far East (Japan), but Gaspar and Gómez Cruz<sup>5</sup> reported the first case in Mexico, in 2004; then, two other cases took place in less than one year, which raised a suspicion that the disease was more common than expected in our region and probably there was an undercount because of its similarity with acute coronary syndrome<sup>1,5</sup>.

## CASO CLÍNICO

A 54-year-old woman with a history of rheumatic fever, which –approximately one year before diagnosis– began with symptoms of heart failure (functional class III according to the New York Heart Association) and palpitations. Atrial fibrillation and severe mitral stenosis were diagnosed, with valve area of 0.9 cm<sup>2</sup> and elevated diastolic gradients. The preoperative screening was indicated, which included a coronary angiography where the coronary arteries without lesions were found, and the other tests were normal.

The surgery was carried out with an anoxic arrest time of 42 minutes and extracorporeal circulation time of 60 minutes, where a bileaflet mechanical prosthetic valve Carbomedics number 27 was implanted, with an uncomplicated transoperative phase. She was moved to the Intensive Care Unit inotropic support of dobutamine mcg/kg/min) and 30 minutes later, she presented a severe low cardiac output, with cardiorespiratory arrest, thus, the usual resuscitation maneuvers were applied, but having no proper response, the reopening of the median longitudinal sternotomy was decided for an internal cardiac massage, and there was observed a left ventricle with hypokinesia of its entire anterior, lateral and apical walls; only the base was shrinking. The inotropic support was not enough to achieve improvement of hemodynamic parameters, therefore, a balloon of intraaortic counterpulsation was placed for support 1:1.

The patient recovered the hemodynamic parameters, although she remained with hypotension and the electrocardiogram showed ST elevation from  $V_4$  to  $V_6$ . The case was collectively discussed, when

considering the acute ischemic event (perioperative infarction) as the most likely cause, there was decided to perform an urgent coronary angiography; but the patient experienced an evident improvement and the procedure was performed 36 hours later, where normal coronary arteries were evidenced (Fig. 1. A & B), as well as normofunctional mitral prosthetic valve (in the fluoroscopy, Fig. 1. C & D), without paravalvular leaks or obstructions when performing the ventriculography, where a significant motility disorder of the entire left ventricle was observed, except its base; this image evidenced what is describe as the Takotsubo syndrome (Fig. 2).

Although the electrical and echocardiographic recovery occurred in the first 72 hours, her definitive recovery was longer, due to the presence of complications such as respiratory distress in need of mechanical artificial ventilation, pneumonia associated with ventilation (required tracheostomy), post-operative mediastinitis and sternal dehiscence; all this resulted in a long hospital stay, but the patient survived and was discharged with good functional capacity, which she has maintained in the external consultation follow-up.

### **COMENTARIOS**

The importance of identifying the Takotsubo syndrome is because it appears similar to an acute myocardial infarction, but with different evolution and prognosis, that is why its treatment is different<sup>5</sup>. Until 2001, all publications in this regard had been from isolated cases or small series, from two to eight patients, but –according to Gaspar and Gómez Cruz<sup>5</sup>–between 2001 and 2002, two large retrospective series from Japan were published: one of 88 patients studied among 1991 and 2000 in 19 hospitals and the other 30, detected between 1983 and 2001 in a single center. These and other recent publications have allowed a better characterization of this syndrome.

There are psychological or physical trigger factors, among which is highlighted the unusual and intense emotional stress, from any cause, vigorous exercise, the surgical procedures or the exacerbation of systemic diseases<sup>1.5</sup>

The Takotsubo syndrome is a reversible, acute cardiomyopathy with the same symptoms of an infarction, elevation of biomarkers and acute ischemic electrocardiographic changes. In imaging studies, an important alteration in the contractility of the entire apical cap is highlighted, with compensatory basal

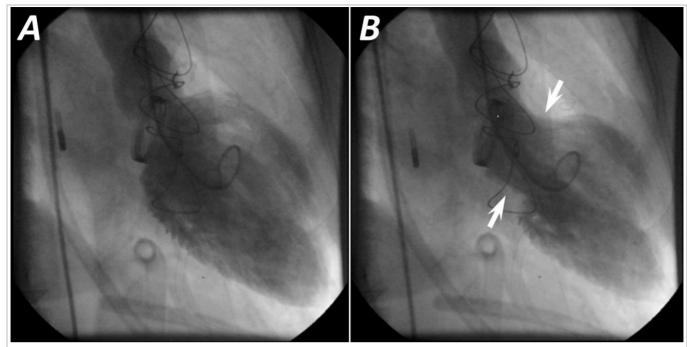


Fig. 2. Left ventriculography A. Diastole. B. Systole (the arrows indicate the contraction of the basal segments).

hypercontractility. In some cases, the systolic function is so depressed that the hemodynamic situation of the patient becomes Killip IV (cardiogenic shock)<sup>6</sup>, as it happened in the case presented here. If the patient overcomes that moment, it is striking the rapid normalization of ventricular function, low elevation of necrosis markers (taking into account the extensive myocardial involvement) and myocardial alteration of several coronary territories, without significant lesions in the corresponding epicardial vessels<sup>6,7</sup>.

The stress trigger, which involves the pathophysiology of the syndrome, has a strong environmental component. However, it is conceivable that some people have genetic predisposition to suffer Takotsubo syndrome. There has been registered the presence or absence of functional polymorphisms in genes such as alpha 1, beta 1 and beta 2 adrenegic receptors, GRK5 and involvement of estrogens<sup>8,9</sup>.

In the primary Takotsubo syndrome, the acute cardiac symptoms are the main reason for seeking medical attention; on the other hand, the secondary takes place in patients hospitalized for another cause, where this associated disease is identified<sup>10</sup>. In these patients, the sudden activation of the sympathetic nervous system or the increase in catecholamines is a complication of the primary medical situation<sup>10,11</sup>.

Few reports of patients with Takotsubo syndrome in postoperative cardiovascular surgery have been found, demonstrating that it is a rare complication. Lorca et al<sup>7</sup> found it in a 68-year-old woman who had been discharged a month ago, after a double mitral and aortic valve replacement, with normal electrocardiogram and echocardiogram at the time of discharge. Attisani et al<sup>12</sup>, in a 77-year-old woman, after a mitral minimally invasive surgery, plus ablative epicardial atrial fibrillation; also the case of Kogan et al<sup>13</sup>, a 62-year-old woman, after an elective surgery of mitral valve replacement and tricuspid annuloplasty. Moreover, Yamane *et al* <sup>14</sup> describe it in a 68year-old woman, five hours after an uncomplicated mitral valve replacement surgery plus tricuspid repair.

Some authors have reported cases of Takotsubo following non-valvular heart surgery, such as aneurysm of the noncoronary sinus of Valsalva with rupture into right atrium<sup>6</sup> and left atrial myxoma<sup>15</sup>; and others, after thoracic extracardiac surgery<sup>16</sup>or extrathoracic surgery<sup>17,18</sup>; but all agree that, stress prior to the surgery, with consequent elevation of endogenous catecholamines and vasoactive drug administration –such as dobutamine–, are all factors involved in the occurrence of this syndrome in the perioperative stage.

# **REFERENCES**

- Morales-Hernández AE, Valencia-López R, Hernández-Salcedo DR, Domínguez-Estrada JM. Síndrome de Takotsubo. Med Int Méx. 2016;32(4): 475-91.
- 2. Peters MN, George P, Irimpen AM. The broken heart syndrome: Takotsubo cardiomyopathy. Trends Cardiovasc Med. 2015;25(4):351-7.
- 3. Kato K, Lyon AR, Ghadri JR, Templin C. Takotsubo syndrome: aetiology, presentation and treatment. Heart. 2017;103(18):1461-9.
- 4. Pelliccia F, Kaski JC, Crea F, Camici PG. Pathophysiology of Takotsubo Syndrome. Circulation. 2017;135(24):2426-41.
- 5. Gaspar J, Gómez Cruz RA. Síndrome Tako-Tsubo (Discinesia antero-apical transitoria): Primer caso descrito en América Latina y revisión de la literatura. Arch Cardiol Méx. 2004;74(3):205-14.
- 6. Chiariello GA, Bruno P, Colizzi C, Crea F, Massetti M. Takotsubo Cardiomyopathy Following Cardiac Surgery. J Card Surg. 2016;31(2):89-95.
- 7. Lorca R, Callejo F, Pun F, Martín M, Corros C, Alperi A, *et al.* Takotsubo syndrome after heart valve surgery. Int J Cardiol. 2015;197:254-6.
- 8. Pizzino G, Bitto A, Crea P, Khandheria B, Vriz O, Carerj S, *et al.* Takotsubo syndrome and estrogen receptor genes: partners in crime? J Cardiovasc Med (Hagerstown). 2017;18(4):268-276.
- 9. Saif MW, Smith M, Maloney A. The First Case of Severe Takotsubo Cardiomyopathy Associated with 5-Fluorouracil in a Patient with Abnormalities of Both Dihydropyrimidine Dehydrogenase (DPYD) and Thymidylate Synthase (TYMS) Genes. Cureus [Internet]. 2016 [citado 18Abr 2019];8(9):e783. Available at: http://doi.org/10.7759/cureus.783
- 10. Núñez-Gil IJ, Almendro-Delia M, Andrés M, Sionis A, Martin A, Bastante T, *et al.* Secondary forms of

- Takotsubo cardiomyopathy: A whole different prognosis. Eur Heart J Acute Cardiovasc Care. 2016;5(4):308-16.
- 11. Chhabra L. Comparison of mortality in primary and secondary Takotsubo cardiomyopathy with severe left ventricular dysfunction. Eur J Heart Fail. 2019;21(8):1046.
- 12. Attisani M, Campanella A, Boffini M, Rinaldi M. Takotsubo cardiomyopathy after minimally invasive mitral valve surgery: clinical case and review. J Heart Valve Dis. 2013;22(5):675-81.
- 13. Kogan A, Ghosh P, Schwammenthal E, Raanani E. Takotsubo syndrome after cardiac surgery. Ann Thorac Surg. 2008;85(4):1439-41.
- 14. Yamane K, Hirose H, Reeves GR, Marhefka GD, Silvestry SC. Left ventricular dysfunction mimicking Takotsubo cardiomyopathy following cardiac surgery. J Heart Valve Dis. 2011;20(4):471-3.
- 15. García-Delgado M, García-Huertas D, Navarrete-Sánchez I, Olivencia-Peña L, Garrido JM. Soporte con oxigenación de membrana extracorpórea en un síndrome de Takotsubo y QT largo tras cirugía cardiaca. Med Intensiva. 2017;41(7):441-3.
- 16. Kinoshita F, Toyokawa G, Tagawa T, Matsubara T, Kozuma Y, Haratake N, *et al.* Takotsubo Cardiomyopathy Developed After Two-stage Surgery for Double Primary Lung Cancer. Anticancer Res. 2018;38(5):2957-60.
- 17. Hammer N, Kühne C, Meixensberger J, Hänsel B, Winkler D. Takotsubo cardiomyopathy An unexpected complication in spine surgery. Int J Surg Case Rep. 2015;6(C):172-4.
- 18. Busse EC, Wiater JM. Perioperative Takotsubo Cardiomyopathy: A Rare Cardiac Complication Following Orthopaedic Surgery: A Case Report. JBJS Case Connect [Internet]. 2015 [citado22 Abr 2019];5(3):e64. Available at:
  - http://doi.org/10.2106/JBJS.CC.N.00215