

DOI: 10.21767/2471-8041.100049

Reverse Takotsubo Cardiomyopathy and Shock Supported by Continuous Hemodiafiltration in a Patient with Anorexia Nervosa

Beppu H, Yoshida Y, Katsuki T and Hinoshita F

Department of Nephrology, National Center for Global Health and Medicine, Japan

Corresponding author: Fumihiko Hinoshita, Head, Department of Nephrology, National Center for Global Health and Medicine, Japan, Tel: +81 3-3202-7181; E-mail: fhinoshi@hosp.ncgm.go.jp**Rec Date:** April 16, 2017, **Acc Date:** April 26, 2017, **Pub Date:** April 28, 2017**Citation:** Beppu H, Yoshida Y, Katsuki T. Reverse Takotsubo Cardiomyopathy and shock supported by continuous hemodiafiltration in a patient with anorexia nervosa. *Med Case Rep*, 3:2.

Abstract

Background: Takotsubo cardiomyopathy (TC) is known as stress-induced cardiomyopathy. It is characterized by relatively benign transient systolic dysfunction of the apical segments of the left ventricle. Reverse Takotsubo cardiomyopathy (RTC) is a rare variant of TC. Here we describe a rare case of RTC and cardiogenic shock in a patient with anorexia nervosa (AN).

Case presentation: A 54-year-old woman with AN showed RTC due to subarachnoid hemorrhage (SAH) stress; renal replacement therapy including continuous hemodiafiltration (CHDF) was successfully performed to maintain her cardiohemodynamic state.

Conclusion: To our knowledge, this is the first documented report showing that renal replacement therapy greatly contributed to the circulation management and recovering kidney dysfunction with anuria in a patient with an eating disorder and RTC.

Keywords: Anorexia nervosa; Reverse Takotsubo cardiomyopathy; Continuous hemodiafiltration; Subarachnoid haemorrhage; Congestive heart failure; Chronic kidney disease

characterized by transient systolic dysfunction of the apical segments of the left ventricle mimicking myocardial infarction in the absence of obstructive coronary artery disease. Reverse Takotsubo cardiomyopathy (RTC) is a rare variant of TC; it is one of four stress-induced cardiomyopathy patterns in which the basal and midventricular segments of the left ventricle are akinetic [2]. According to the International Takotsubo Registry (ITR) [3], RTC was found in 2.2% of 1,750 cases with stress-induced cardiomyopathy. The mechanism of TC is still controversial, but it is considered that TC is triggered by emotional or physical stress [4-7]. We encountered a very interesting patient with anorexia nervosa (AN) who developed RTC and shock following subarachnoid hemorrhage (SAH) and recovered with the support of continuous hemodiafiltration (CHDF). To the best of our knowledge, this is the first case report of RTC with shock treated by CHDF in a patient with AN.

Case Report

A 54-year-old woman was admitted to the Department of Nephrology, National Center for Global Health and Medicine (NCGM) Hospital, for the treatment of liver dysfunction. At the age of 38, she first developed AN (binge eating/purging type). She was found to have chronic kidney disease (CKD) at the age of 53 due to chronic hypokalemia and low renal blood flow. Then, she started outpatient visits at Department of Nephrology, NCGM Hospital, and routinely has been administered an intravenous drip infusion to prevent dehydration. In October of 2016, she was admitted to our hospital because of liver dysfunction caused by abnormalities in intravenous amino acid administration.

On physical examination, she appeared poorly nourished. Her height was 157 cm and body weight was 24.2 kg. Her pulse rate was 52 bpm and blood pressure was 89/53 mmHg on admission. Complete blood counts and coagulation activity test revealed; white blood cell count 3,120 /mm³; hemoglobin 9.8 g/dl; platelet count 207,000 /mm³; prothrombin time 11.9 s; aPTT, 26.3 sec; and fibrinogen 179.3 mg/dl. Biochemical examinations showed; albumin 3.4 g/dl; total bilirubin 1.4 mg/dl; serum aspartate aminotransferase 260 U/l; alanine aminotransferase 204 U/l; lactate dehydrogenase 449 U/l; urea nitrogen 78.4 mg/dl; creatinine 4.5 mg/dl; sodium 135 mmol/l; potassium 3.4 mmol/l; calcium 8.0 mg/dl; phosphorus 4.0

Abbreviations

AN: Anorexia Nervosa, TC: Takotsubo Cardiomyopathy, SAH: Subarachnoid Hemorrhage, RTC: Reverse TC, ITR: International Takotsubo Registry, HD: Hemodialysis, CHDF: Continuous Hemodiafiltration, CKD: Chronic Kidney Disease, ECG: Electrocardiogram, CT: Computed Tomography, CTA: Computed Tomography Angiography, CHF: Congestive Heart Failure, TTE: Transthoracic Echocardiography

Introduction

Takotsubo cardiomyopathy (TC), first described in Japan in 1990 [1], is known as stress-induced cardiomyopathy. It is

mg/dl. Kidney dysfunction had been previously identified, but it further progressed. The electrocardiogram (ECG) showed no ST depression, but low voltage (**Figure 1**).



Figure 1 (A) The electrocardiogram (ECG) on admission, (B) ECG upon transfer to the medical ICU.

On day 7 of admission, she presented with severe anemia (hemoglobin 5.5 g/dl) and an increase in the blood urea nitrogen/creatinine ratio (128.7/3.72 mg/dl). Diffuse gastric oozing was identified by an upper endoscopy, and treated with clipping. No other apparent source of bleeding was found. Gastric oozing was considered to be caused by vascular wall damage with malnutrition. Meanwhile, uremic symptoms such as disturbance of consciousness and severe general weakness appeared. She underwent hemodialysis (HD) 3 times, resulting in gradual clinical improvement (**Figure 2**).

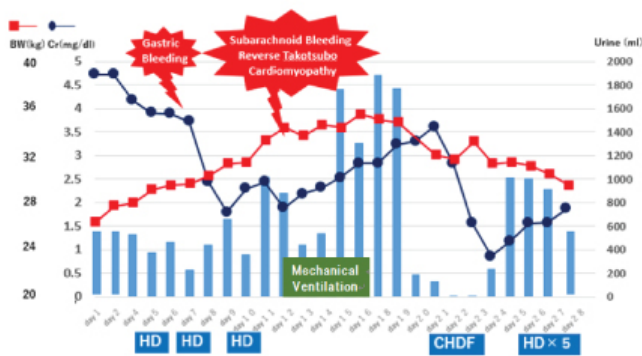


Figure 2 Clinical course HD: Hemodialysis, CHDF: Continuous Hemodiafiltration, Cr: Creatinine, BW: Body Weight.

On day 12 of admission, however, she became more restless and aphasic, and SAH was found by computed tomography (CT) scan (**Figure 3**).

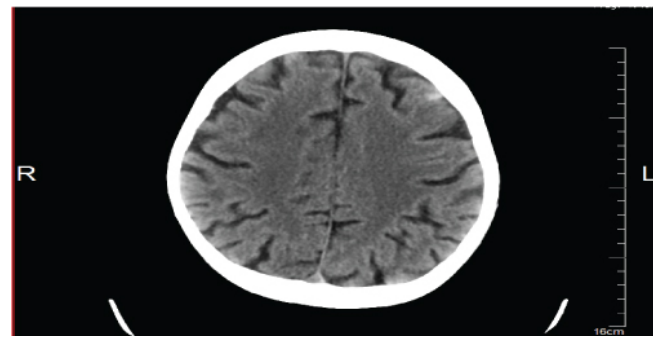


Figure 3 Left frontal subarachnoid hemorrhage (SAH).

We consulted a neurosurgeon. CT angiography (CTA) and MRI were performed, however no responsible or causative vessels were identified and conservative therapy was simply continued. On that evening, the patient suddenly became dyspneic and the chest X-ray revealed newly developed bilateral pleural effusion. Arterial blood gas analysis revealed hypoxia (pH 7.483, PaO₂ 59.4 mmHg, PCO₂ 30.1 mmHg, HCO₃⁻ 22.3 mmol/l, in 15L reservoir mask). She was intubated and transferred to the ICU. ECG showed ST elevation in the anteroseptal leads (V1-V3) (**Figure 1**).

Troponin T was elevated at 1.953 µg/l, and BNP was elevated at 3,666 pg/ml. We consulted a cardiologist, and a bedside echocardiogram was performed to disclose an ejection fraction of 15% to 20%, except for the apical segment which was hyperkinetic (**Figure 4**).

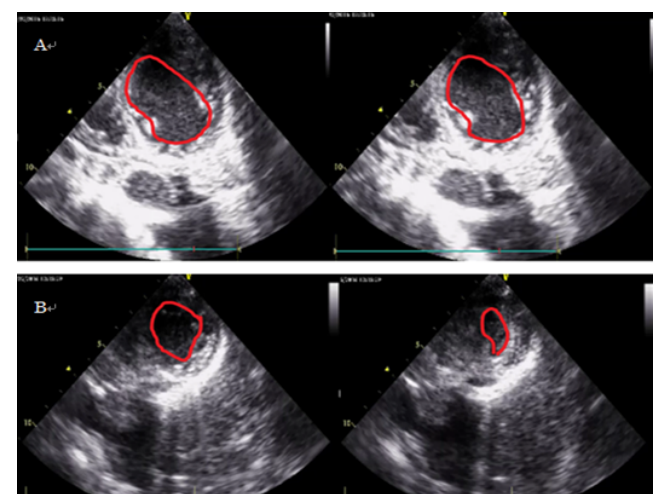


Figure 4 Transthoracic echocardiogram (TTE) upon transferto the medical ICU, (A) Short axis image of basal segment which was hypokinetic, (B) Short axis image of apical segment which was hyperkinetic.

Discussion

Taken together, RTC was diagnosed. Furosemide injection was started and she was extubated 5 days later. However, the

kidney dysfunction rapidly progressed (from creatinine levels 2.53 to 3.66 mg/dl), mainly due to low cardiac function, and she became anuric. Then, CHDF was performed for 3 consecutive days. After discontinuation of CHDF, HD was conducted for water removal 5 times in total. A repeat transthoracic echocardiography (TTE) 7 days later revealed improved ejection fraction, and her serum creatinine level improved to the level of before hospitalization (**Figure 2**).

A dysmorphic girl (**Figure 1**) was referred at the age of four years (early April 2016) because of skin abnormality and delayed speech. Her growth was adequate (height, 103 cm; weight, 23 kg).

We present a 54-year-old female with AN who suffered severe heart failure with cardiogenic shock caused by RTC, requiring CHDF. RTC is much less frequently found compared to apical or classic TC. There have been some case reports demonstrating other types of TC with AN [3], but, to our knowledge, only one report has discussed RTC in an anorexic female patient [8]. In that report, the patient could recover from RTC and heart failure with only conservative therapy. In the present case, however, kidney dysfunction reached the end stage with anuria due to severe congestive heart failure (CHF) which required CHDF and HD.

TC is generally classified into four types based on the involvement of the left ventricle: classic type, reverse type, mid-ventricular type, and localized type [3]. Though RTC has been said to occur in younger patients and recover earlier than other subtypes [9], the clinical course of this case was so severe that temporary mechanical ventilation and renal replacement therapy, including CHDF, were needed. We hypothesized some reasons for this difference. First, she had CKD and fluid management was more difficult than for patients without CKD. Second, it is said that substantial weight loss in AN is accompanied by atrophy of cardiac muscle, decreased cardiac mass, reduced ventricular wall thickness and cardiac chamber volumes [10-12], so CHF could readily and more seriously develop easily in the present case. Third, the physical damage or malinfluence by RTC, gastric bleeding and SAH can be critical on their own. It should be remembered that RTC is not always a mild disease with better prognosis; though rare, it triggers life-threatening CHF and cardiogenic shock with severe kidney dysfunction in such malnourished and bleeding patients.

Patients with RTC have higher prevalence of triggering stress, either physical or emotional, compared to their apical counterparts [13]. Elikowski et al. previously reported 5 cases with RTC caused by intracranial hemorrhage [14]. They indicated a need for further search of the echocardiographic variants of TC if the ECG abnormalities are found in subjects with intracranial hemorrhage. In the present case, gastric bleeding followed by SAH induced great physical stress. In addition, she suffered from serious malnutrition causing liver dysfunction and CKD. CKD is a great risk to induce cardiovascular diseases and easily trigger CHF. Persistent AN for a long period with serious malnutrition is also considered to damage myocardial cells [10-12]. Therefore, there is great possibility that serious RTC might occur in patients with AN

and CKD if some other risk such as intracranial hemorrhage overlaps these diseases.

It is well known that long-term eating disorder or AN can often induce CKD mainly due to chronic hypokalemia and low renal blood flow with dehydration as well as probable interstitial nephritis [15,16]. Silverberg et al. previously suggested the clinical concept or entity of cardio-renal anemia syndrome [17]. Patients with AN and CKD literally get into this cardio-renal anemia syndrome particularly if serious bleeding might occur.

In the present case CHDF followed by HD was effective to treat CHF and the acutely exacerbated CKD. As far as we know, CHDF has never been used to treat CHF and cardiogenic shock caused by RTC in a patient with AN. We think that CHDF might be a good and moderate treatment to support AN patients with RTC and shock. In such patients cardiohemodynamics are unstable with hypotension, and massive infusion of diuretics and regular HD would be very risky. Therefore, CHDF is recommended when regular conservative treatment for CHF is not effective in such patients. Also, intensive care and mechanical ventilation might be helpful. Papanikolaou et al. suggested another novel approach [18]. They tried early co-administration of esmolol and levosimendan in a patient with life-threatening secondary RTC with cardiogenic shock caused by post-caesarean section hemorrhage, and found the co-administration to be effective and safe. This approach also remains to be re-evaluated in those life-threatening patients in the future.

Conclusion

Conclusively, we encountered a life-threatening RTC with shock in a female patient with AN. In general, TC has been considered to be a transient cardiac attack which can be recovered in a few weeks in most of the cases. We suggest, however, that there are a few cases of serious RTC which need rapid diagnosis and active or drastic measures such as CHDF in addition to every regular supportive therapy for CHF and renal failure. Considering the vulnerability to both physical and emotional stress, it is also important to perform ECG and TTE to quickly detect stress-induced cardiomyopathy when anorexic patients complain of chest pain, dyspnea or hypotension.

Declarations

Availability of data and materials

The datasets analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

All authors managed the patient. BH and HF have been involved in drafting the manuscript and revising it critically. All authors approved the final version to be published.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

We have not yet obtained written informed consent but are intended to at the outpatient visit. No individual information can never be identified in this case report because we described this manuscript, paying attention to this serious problem.

Ethics approval and consent to participate

This case report is not an interventional trial with great risk, but a retrospective clinical report without giving any specific damage or unfavorable result to the patient. Therefore, it's unnecessary to be approved by the Ethics Committee.

References

1. Sato HTH, Uchida T, Dote K, Ishihara M (1990) Tako-tsubo-like left ventricular dysfunction due to multi-vessel coronary spasm. Tokyo: Kagakuhyoronsh publishing P: 56-64.
2. Haghi D, Papavassiliu T, Flüchter S, Kaden JJ, Pörner T, et al. (2006) Variant form of the acute apical ballooning syndrome (takotsubo cardiomyopathy): observations on a novel entity. *Heart* 92: 392-394.
3. Templin C, Ghadri JR, Diekmann J, Napp LC (2015) Clinical features and outcomes of Takotsubo (Stress) cardiomyopathy. *N Engl J Med* 373: 929-938.
4. Wittstein IS, Thiemann DR, Lima JA, Baughman KL, Schulman SP, et al. (2005) Neurohumoral features of myocardial stunning due to sudden emotional stress. *N Engl J Med* 352: 539-548.
5. Nef HM, Möllmann H, Elsässer A (2007) Tako-tsubo cardiomyopathy (apical ballooning). *Heart* 93: 1309-1315.
6. Gianni M, Dentali F, Grandi AM, Sumner G, Hiralal R, et al. (2006) Apical ballooning syndrome or takotsubo cardiomyopathy: A systematic review. *Eur Heart J* 27: 1523-1529.
7. Ako J, Sudhir K, Farouque O, Honda Y, Fitzgerald PJ (2006) Transient left ventricular dysfunction under severe stress: Brain-heart relationship revisited. *Am J Med* 119: 10-17.
8. Tagami T, Mertens A, Rothschild D, Chowdhury P (2017) A case of reverse takotsubo cardiomyopathy caused by an eating disorder. *J Cardiol Cases* 15: 77-79.
9. Ramaraj R, Movahed MR (2010) Reverse or inverted takotsubo cardiomyopathy (reverse left ventricular apical ballooning syndrome) presents at a younger age compared with the mid or apical variant and is always associated with triggering stress. *Congest Heart Fail* 16: 284-286.
10. Kraeft JJ, Uppot RN, Heffess AM (2013) Imaging findings in eating disorders. *Am J Roentgenol* 200: W328.
11. Oflaz S, Yucel B, Oz F, Sahin D, Ozturk N, et al. (2013) Assessment of myocardial damage by cardiac MRI in patients with anorexia nervosa. *Int J Eat Disord* 46: 862.
12. Escudero CA, Potts JE, Lam PY, De Souza AM, Mugford GJ, et al. (2016) An echocardiographic study of left ventricular size and cardiac function in adolescent females with anorexia nervosa. *Eur Eat Disord Rev* 24: 26-33.
13. Ghadri JR, Cammann VL, Napp LC, Jurisic S, Diekmann J, et al. (2016) International Takotsubo (InterTAK) Registry. Differences in the clinical profile and outcomes of typical and atypical Takotsubo syndrome: Data from the International Takotsubo Registry. *JAMA Cardiol* 1: 335-340.
14. Elikowski W, Małek-Elikowska M, Kudliński B, Skrzywanek P, Smól S, et al. (2016) ECG pattern in reverse takotsubo cardiomyopathy demonstrated in 5 cases with intracranial hemorrhage. *Pol Merkur Lekarski* 41: 136-140.
15. Bouquegneau A, Dubois BE, Krzesinski JM, Delanaye P (2012) Anorexia nervosa and the kidney. *A J Kidney Dis* 60: 299-307.
16. Zipfel S, Löwe B, Reas DL, Deter HC, Herzog W, et al. (2000) Long-term prognosis in anorexia nervosa: Lessons from a 21-year follow-up study. *The Lancet* 355: 721-722.
17. Silverberg DS, Wexler D, Blum M, Iaina A (2003) The cardio-renal anaemia syndrome: Does it exist? *Clin Nephrol* 8: 7-12.
18. Papanikolaou J, Makris D, Tsolaki V, Spathoulas K, Zakyntinos E (2016) Post-partum hemorrhage complicated by reverse-Takotsubo cardiogenic shock: A novel therapeutic approach. *Am J Emerg Med* S0735-6757.