American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2021; 22: e930484
DOI: 10.12659/AJCR.930484

Received: 2020.12.14
Accepted: 2021.02.24
Available online: 2021.03.19
Published: 2021.04.24

Inverted Takotsubo Cardiomyopathy as an Early Complication After Liver Transplantation

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Female, 49-year-old
Final Diagnosis: Takotsubo cardiomyopathy
Symptoms: Dynpnea
Medication: —
Clinical Procedure: —
Specialty: Anatomy • Critical Care Medicine • General and Internal Medicine • Pathology

Objective: Rare disease

Background: Takotsubo cardiomyopathy (TTC) is a cardiac syndrome characterized by transient left ventricle (LV) dysfunction, typically showing apical ballooning due to apical akinesis with preserved basal segment contractility. The inverted form is very uncommon and is characterized by basal segment hypokinesis or akinesis and normal LV apical segment contractility.

Case Report: We describe the case of a 49-year-old woman who developed inverted TTC after orthotopic liver transplantation. On day 1 (D1), dyspnea and oliguria suddenly appeared. A chest X-ray showed pulmonary edema, and echocardiography showed severe systolic LV dysfunction with an estimated ejection fraction of approximately 25% and akinesis of basal and midventricular LV segments, normal apical segment contractility, and mild mitral regurgitation. Elevated troponin T, creatine kinase-MB, and N-terminal pro B-type natriuretic peptide were found in the blood sample. Suspected inverted takotsubo cardiomyopathy was confirmed by left ventriculography, with normal apical part motion, akinesis in the other LV parts, and negative coronary angiography. The echocardiographic findings returned to normal on D14, and the patient was discharged from the hospital on D19 with normal LV motion and an ejection fraction of 65%. The transplanted liver function was excellent.

Conclusions: Organ transplantation is connected with a great emotional stress because the patient's life depends on the death of another person. Therefore, we have to think about the possibility of stress cardiomyopathy even after liver transplantation, because early diagnosis and treatment can be life-saving for the patient. To our knowledge, this is the first described case of inverted takotsubo cardiomyopathy after liver transplantation.

Keywords: Female • Liver Diseases, Alcoholic • Stress, Psychological • Takotsubo Cardiomyopathy

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/930484

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Background

Takotsubo cardiomyopathy (TTC) is a cardiac syndrome characterized by transient left ventricle (LV) dysfunction, typically showing apical ballooning due to apical akinesis with preserved basal segment contractility. The inverted form is very uncommon and is characterized by basal segment hypokinesis or akinesis and normal LV apical segment contractility.

Case Report

A 49-year-old woman underwent liver transplantation in July 2019 for end-stage alcoholic liver cirrhosis. The patient has been monitored for hepatopathy since 2007. From the beginning of 2017, icterus and ascites began to appear, and in March 2019 she underwent severe decompensation with the need for an ascites puncture. Supplemental nutrition was also introduced at that time for protein-caloric malnutrition leading to the patient being underweight, with a body mass index of 15 kg/m^2. The patient had no comorbidities other than those associated with the primary disease, and no history of coronary artery disease. Transthoracic echocardiography prior to transplantation demonstrated normal left ventricular size and function (ejection fraction was 65%). The intraoperative course was uneventful. The patient did not require inotropic support during the procedure and had no hemodynamic instability. She was extubated 2.5 h after surgery. On day 1 (D1), dyspnea and oliguria suddenly appeared. A chest X-ray showed pulmonary edema (**Figure 1**), and echocardiography showed severe systolic LV dysfunction with an estimated ejection fraction of approximately 25% and akinesis of basal and midventricular LV segments, normal apical segment contractility, and mild mitral regurgitation. Elevated troponin T 219 ng/L (normal range 0-14), creatine kinase-MB 0.53 µkat/L (normal range 0-0.42), and N-terminal pro B-type natriuretic peptide >35.000 ng/L (normal range 0-300) were found in the blood sample. A 12-lead electrocardiogram demonstrated sinus tachycardia of 130 beats/min, with no ischemic changes (**Figure 2**). Suspected inverted takotsubo cardiomyopathy was confirmed by left ventriculography (**Figure 3**), with normal apical part motion, akinesis in the other LV parts, and negative coronary angiography. During the examination, the patient's dyspnea became worse and reintubation was therefore performed. The patient required inotropic support (dobutamine and levosimendan) and high doses of diuretics. Her condition improved gradually, the laboratory signs of heart failure decreased, and the echocardiographic findings returned to normal on D14. She was discharged from the hospital on D19 with normal LV motion and an ejection fraction of 65%. The transplanted liver function was excellent, with normal liver test values. More than 1 year after liver transplantation, the patient has good liver and heart function and is living a full life.

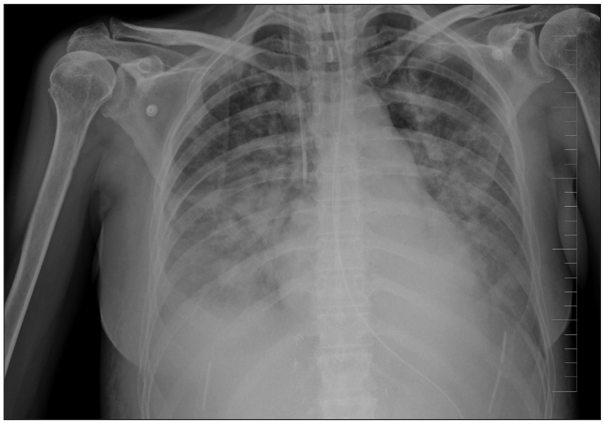

Figure 1. Chest X-ray of the patient showing pulmonary edema.

Discussion

Stress cardiomyopathy was first described in 1990 by Sato et al [1] as an acute cardiac disease mimicking acute coronary syndrome (ACS). It is characterized by a sudden onset of chest pain or dyspnea, electrocardiographic changes, mild cardio-specific marker positivity, and the development of a typically apical LV kinetic disorder with preserved base kinetics and no significant coronary artery involvement. The disease is synonymous with TTC, according to the shape of the affected left ventricle resembling an octopus trap (ie, takotsubo) in Japan [1]. In addition to this typical form, 3 other types of LV involvement have been described: inverted, midventricle, and localized. Kawai et al [2] described the frequency of classic, inverted, and midcavitary types as 67%, 23%, and 10%, respectively. Often (but not necessarily), there is a stress (emotional or physical) trigger, absence of significant coronary involvement, absence of acute rupture of plaque or thrombosis, and new electrocardiographic changes (ST elevation, T-wave inversion, or both), mild elevation of troponin in the heart, pheochromocytoma, myocarditis or hypertrophic cardiomyopathy, or intracranial injury and bleeding [2]. A triggering stressor is usually present in the classic variant, but not usually present in patients with the inverted form [3]. Typically, this syndrome occurs in women. The stressogenic stimulus in our patient was a liver transplantation. The stress etiology of TTC is illustrated by a higher incidence of this disease during the COVID-19 pandemic. In a recent publication, Jabri et al [4] reported a 7.8% incidence of stress cardiomyopathy during the COVID-19 pandemic, which was significantly increased compared with prepandemic incidences ranging from 1.5% to 1.8%. Stress cardiomyopathy mainly affects postmenopausal women, which was the case for our patient, who had secondary amenorrhea. The more frequent disease occurrence is explained by the decreased estrogen levels in this population. Decreased estrogen is associated with endothelial dysfunction and a change in the heart's reactivity to catecholamines [5]. The disease is typically preceded by greater physical, mental, or perioperative stress,

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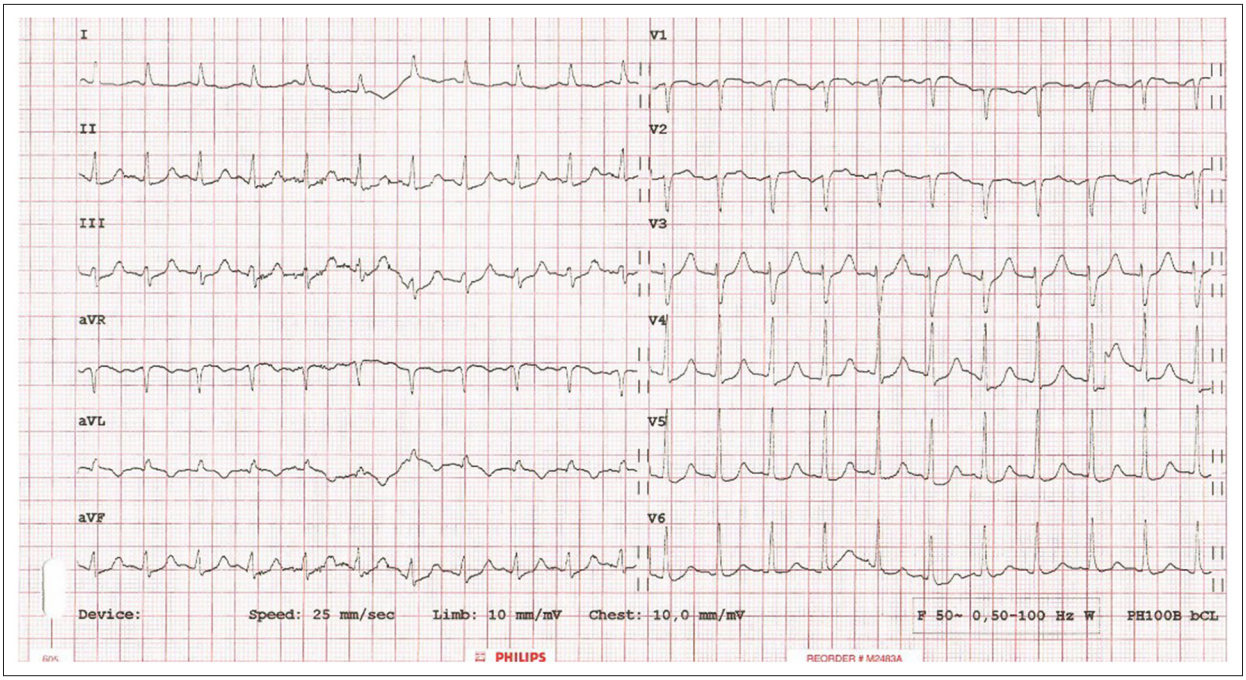

Figure 2. The patient's electrocardiogram showing sinus tachycardia and nonspecific changes.

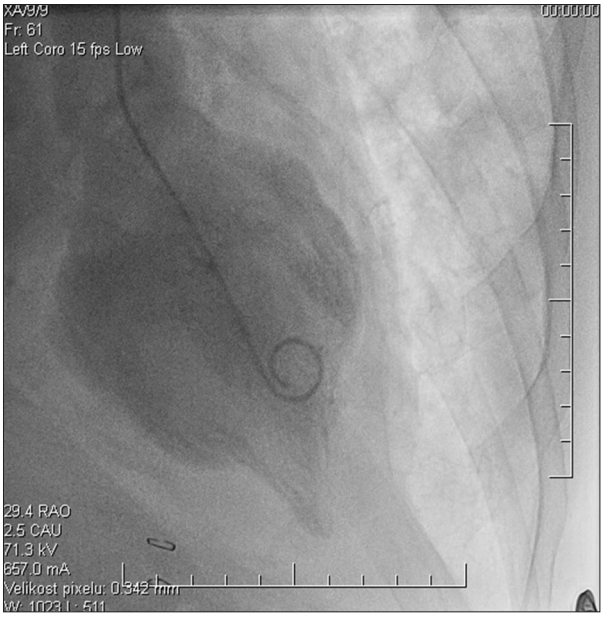

Figure 3. Left ventriculography during systole showed normal apical part motion, with akinesis in the other parts of the left ventricle.

and its estimated prevalence is around 1.2% that of ACS [5-7]. All forms of TTC have better short- and long-term prognosis than ACS. The diagnosis of stress cardiomyopathy requires ruling out ACS and other forms of ischemic heart disease. Cases involving the right ventricle and basal LV part have also been described [7]. The unambiguous etiopathogenesis of the disease is not clear. Three etiopathogenetic concepts are currently being considered: catecholamine cardiotoxicity, microcirculatory disorders, and neurogenic myocardial stunning. The disease is associated with markedly elevated catecholamine levels, and histological changes are similar to the changes in myocardial involvement in a pheochromocytoma [2,7]. Neurogenic stunning suggests changes similar those that occur in subarachnoid hemorrhage [2,7]. ST segment elevation is present in about 68% of patients, and T-wave diffuse inversion is seen in up to 97%. QT prolongation is typical [8]. The similarity of the diagnosis of TTC to ACS is very narrow. Strictly distinguishing between them may be impossible in certain situations, even with the use of intravascular ultrasound. Microcirculation damage is similar in both pathologies, but in stress cardiomyopathy there is no scar development. Stress cardiomyopathy could be regarded as an "atypical" form of ACS. The coexistence of stress cardiomyopathy and coronary heart disease is reported in about 5% of cases [7]. Since 2003, considerable efforts have been undertaken to identify criteria to facilitate the diagnostics. The best known is the recommendation of the Mayo Clinic [9], which was first published in 2004 and further modified in 2008 and which remains the most applied in clinical practice. The International Takotsubo Registry developed the so-called diagnostic criteria InterTAK [10]. They are highly specific, however; even the lowest score does not mean 100% exclusion, while the maximum number of points does not confirm the diagnosis. InterTAK criteria contain 7 items rated at different points: female sex, emotional and physical stress, psychiatric or neurological disorder, prolonged QTc interval, and the absence of ST segment depressions.

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Treatment for TTC is empirical only. Although the clinical presentation, outcome, and management are similar between apical and inverted TTC, a triggering stressor is often present in all types of TTC, with some differences between the inverted type and other variants [11]. Patients with the inverted form tend to be younger and have lower prevalence of dyspnea, T-wave inversion, and acute mitral regurgitation [2,3,]. Therefore, recommended treatment for pulmonary edema includes diuretics, angiotensin-converting enzyme inhibitors, oxygen, and nitrates. In cases of resistant pulmonary edema or cardiogenic shock, artificial lung ventilation and the use of intra-aortic balloon counter-pulsation are indicated. The prognosis of these diseases in patients who are already hospitalized is favorable, and the reported mortality is below 1% [11]. The published cases of death are associated with LV free wall rupture, cardiogenic shock, or other, often extracardiac complications [12]. The recurrence rate is approximately 10% [13].

Conclusions

We described a rare case of inverted TTC in a 49-year-old woman after liver transplantation. Organ transplantation is understandably connected with intense emotional stress because the patient's life depends on the death of another person. Therefore, we have to think about the possibility of stress cardiomyopathy even after liver transplantation because early diagnosis and treatment can be life-saving for the patient.

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