Stuck in the upside down tackotsubo cardiomyopathy: A case of inverted tackotsubo cardiomyopathy during exploratory thoracotomy.

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Abstract

Background: Takotsubo cardiomyopathy is characterized by relatively benign transient systolic dysfunction of the apical segments of the left ventricle. Inverted (Upside down) Takotsubo Cardiomyopathy (ITC) is a rare variant of TC. ITC is characterized by reversible regional wall motion abnormalities in the basilar segment of the left ventricle. Although it shares some pathogenic mechanisms with its more common variant, TC, differences exist in terms of echocardiographic features, demographics, clinical manifestations, laboratory features and prognosis. Here we pre-sent a case of intra-operative ITC.

Case Summary: We report a case of ITC in a 44-year-old man who presented for an elective biopsy from mediastinal mass and suffered a peri-procedural cardiac arrest. He came to the hos-pital with dyspnea on exertion that had lasted for a few months but became exacerbated in the last three weeks. In the operating room; after uncomplicated induction of anaesthesia before the surgery, the patient went into pulseless electrical activity. The patient was resuscitated and maintained on supportive therapy. He was taken urgently for cardiac catheterization, which showed normal coronary arteries with akinetic basal and inferior walls and hyperkinesis of the apex. His ejection fraction decreased to 30% from 70% at baseline. He was placed on an appropriate heart failure regimen. An echocardiogram after two weeks showed complete resolution of cardiac dysfunction with symptoms improvement. With this normal coronary angiogram and transient systolic dysfunction of the mid and basal segments of the left ventricle; he was diagnosed with inverted (Upside down) TC.

Conclusion: We offer this case as a reminder that stress during the surgical procedure should be considered one of the differential diagnosis of aetiology in patients presenting with TC. With supportive care and initiating treatment in a timely fashion, the long-term prognosis will be good in the vast majority of patients.

Keywords: Case report, Inverted takotsubo, Cardiomyopathy, Peri-operative.

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Introduction

Stress-induced cardiomyopathy or Takotsubo Cardiomyopathy (TC) is a transient left ventricular myocardial stunning and dysfunction in the absence of coronary artery stenosis or acute plaque rupture [1]. In most cases of TC, the cardiac wall motion abnormality does not follow a single coronary artery territory. On the other hand, Inverted TC (ITC) has been recognized as a variant with preserved apical wall function [2]. The mechanism of all types of TC considers to be triggered by extreme and sudden emotional or physical stressful events, and its symptoms mimic myocardial infarction [3]. Symptoms often improve quickly as the initially impaired cardiac function is usually restored within days or weeks [4]. A case of ITC following surgery seems to be rare [5]. We reported a scenario with mediastinal mass who developed ITC when underwent exploratory thoracotomy.

Case Summary

We present a case of inverted TC in a 44-year-old man occurring during exploratory operation. He came to our emergency department with a chief complaint of dyspnea on exertion that had lasted for a few months but became exacerbated in the last three weeks. Other relative symptoms included mild non-specific chest pain on exertion, fever, diaphoresis, and dysphagia. He noticed weight loss in the past four months. He was a heavy smoker but had no medical history of hyper-tension, diabetes mellitus, coronary ischemia, dysrhythmias or history of drug abuse. The patient was diagnosed with mediastinal mass after a contrast-enhanced computed tomography scan of his chest. He was admitted to do exploratory thoracotomy for biopsy. On admission, he had a blood pressure of 120/70 mmHg, heart rate of 80 Beats Per Minute (bpm) and oxygen saturation of 98% on room air. He was brought to the operating room. Electrocardiogram was remarkable only for sinus tachycardia. After uncomplicated induction of anaesthesia before the surgery, sinus bradycardia associated with profound hypotension (70/40 mmHg) ensued. Despite the administration of epinephrine and atropine, the patient's condition worsened to cardiovascular collapse. Cardiopulmonary Resuscitation (CPR) was initiated, and Return Of Spontaneous Circulation (ROSC) was achieved within the 60 s. After ROSC, his vital signs were notable for sinus tachycardia with systolic blood pressure less than 90 mmHg despite continuous infusions of high-dose norepinephrine (3 μg/Kg/min) and dopamine (20 μg/kg/min) for hemodynamic support. Because of worsening hypoxemia, high flow nasal cannula oxygen was initiated. The surgical procedure was

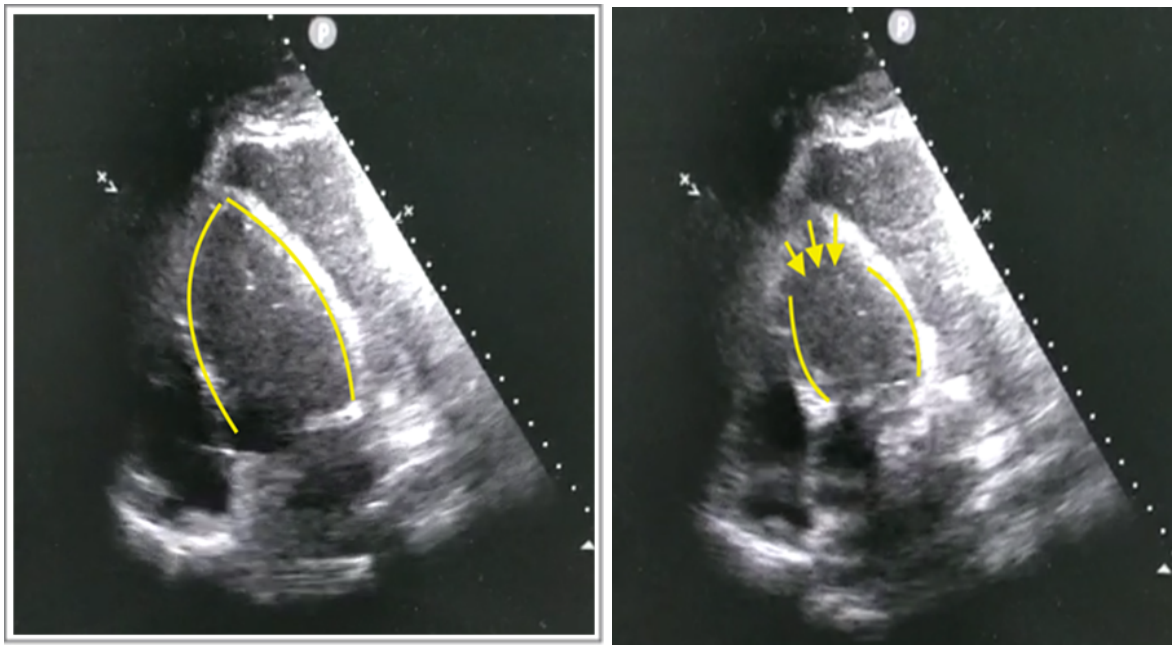

Figure 1. *Transthoracic echocardiogram (apical 4 chamber view) shows hypokinesia of basal and midventricular segments (lines) and hyperkinesia of the apical segments (arrows) in an inverted-Takotsubo type pattern;* ***(A)*** *Shows end of diastole and;* ***(B)*** *shows end of systole.*

finished in a short time with minimal blood loss, and the patient was transferred to the coronary care unit. The electrocardiogram post-ROSC revealed sinus tachycardia with new T-wave inversions in the precordial leads. The echocardiography showed normal contracting apex and diffusely hypokinesia of the rest of the left ventricle and depressed Left Ventricular Ejection Fraction (LVEF); EF decreased to 30% from 70% five days before the surgical procedure. The function and size of the right ventricular were normal with no pericardial effusion. The level of Creatine Kinase MB Fraction (CK-MB) was 14.6 (reference range: <3.8) and repeated troponin I measurement levels were elevated and peaked at 3665.2 pg/ml (reference range: <26.2 pg/ml). Given the clinical picture suspected non-ST elevation myocardial infarction, the patient was taken urgently for cardiac catheterization. The coronary anatomy was notable for no obstructive coronary lesions to explain his depressed EF. Left ventriculography revealed marked hypokinesia of the basal and mid inferior and the anterolateral walls, with the apex being hyperdynamic, and severely reduced EF of 30% as shown in the Figure 1. By the end of postoperative day 1, the patient was no longer in cardiogenic shock. Therefore, inotropic and mechanical supports were weaned off, and he was successfully extubated. Therapy with escalating doses of diuretics, β-blocker and an angiotensin-converting-enzyme inhibitor was initiated. After five days, the patient was discharged after a noticeable improvement of his general condition with a persistent LVEF of 30%. An echocardiogram was repeated after two weeks that revealed complete resolution of cardiac dysfunction, and the patient denied any symptoms of heart failure. His EF normalized to 70% without regional wall motion abnormalities. With normal coronary angiogram and this transient systolic dysfunction of the mid and basal segments of the left ventricle with sparing the apex; he was diagnosed with inverted TC. The patient is currently being followed up at the oncology outpatient department as he was diagnosed by open biopsy as a case of mediastinal large B-cell Lymphoma, Stage II.

Discussion

Herein, we report a case of an inverted pattern of TC in a young man occurring during exploratory thoracotomy. In TC, there is apical ballooning due to hyperkinetic basal and inferior wall motion and apical hypokinesis as shown in the Figure 2. In an inverted type of TC; as in our patient, the apex is hypercontractile in contrast to the base, which is akinetic or hypokinetic. Also, the pattern of myocardial dysfunction in TC usually does not follow any single coronary artery territory. It instead tends to be limited to horizontal areas along the longitudinal axis of the left ventricle. The reason for this distribution is not yet well understood, but variations in the distribution, amount and sensibility of beta-1 and beta-2 adrenoceptors in the myocardium may play an important role. Those hypokinetic areas are the areas with a higher density of adrenergic receptors [6]. Adrenoreceptor concentration is highest in the apex compared with the base in postmenopausal women, which explains the occurrence of the apical variant in older women [7]. Others hypothesized that the presentation of inverted TC at a young age might be due to the abundance of adrenoreceptors at the base compared to the apex [8]. Our patient was young, and this may be the reason why he developed the inverted variant of TC. Our case is not a common, according to the International Takotsubo Registry (ITR), which included 1750 patients with stress cardiomyopathy, who were divided into 4 types; apical akinesia and basal hyperkinesia-82%, basal akinesia and apical hyperkinesia-2.2%, mid-ventricular-14.6% and focal-1.5% in ITR [9]. Cases of perioperative inverted TC are less described in the literature. Concordant with us, in 2010, Sahng Lee presented two unique case reports of inverted TC; one of them was postoperative [10]. It is presumed that severe emotional and physical stress of the surgery triggered this state. The stressful event in cases of TC, usually emotional, but can be induced by physical factors, illness, medical/surgical procedures or sympathomimetic drugs [3]. Activation of sympathetic and

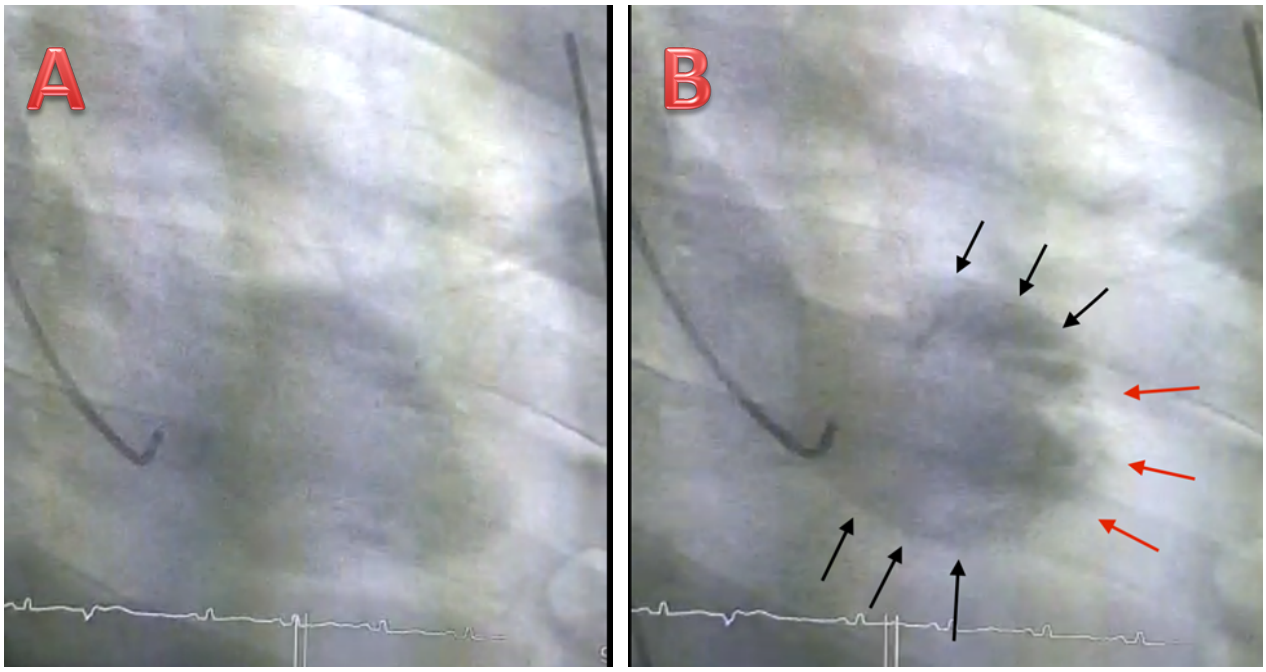

Figure 2. *Left ventricle angiography confirms ballooning of basal and midventricular sections (black arrows) and hypercontractile apex (red arrows);* ***(A)*** *Shows end of diastole and;* ***(B)*** *shows end of systole.*

adrenomedullary hormonal systems resulting in an adrenergic storm have been suggested as the mechanism for the occurrence of TC [11]. High levels of catecholamines are toxic to cells via generation of free radicals and induction of apoptosis [12]. An alternative possibility is sympathetically mediated microvascular spasm causing abnormal coronary flow in the absence of obstructive disease [13]. Interestingly, it is noted that patients with inverted TC have significantly higher levels of Creatine Kinase MB fraction (CK-MB) and troponins. In our case, the level of CK-MB and repeated troponin I measurement levels were elevated, making cardiac catheterization a necessary step in ruling out acute coronary syndrome. The possible explanation of an elevated troponin level is that the inverted TC variant has a greater extent of affected myocardium compared to the classic form where a smaller apical segment is involved [14]. In the vast majority of cases of TC, normalization of the ejection fraction, and complete recovery of the patient happened quickly [15] that matched with our case scenario. Our patient developed cardiac arrest, followed by cardiogenic shock. According to ITR, chest pain, shortness of breath and syncope are the most common presenting symptoms. Some patients present with signs and symptoms of heart failure and up to 10% of patients might present with signs and symptoms characteristic of cardiogenic shock [9]. By open biopsy, our case was diagnoses as having mediastinal large B-cell Lymphoma. Although cancer patients have much comorbidity, TC is rare among those patients. On the other hand, cancer may play a role as a trigger of TC. Vejpongsa et al. reported that; among the cancer-related TC cases, lymph proliferative disorder (30%), gastrointestinal cancer (15%), and lung cancer (12.5%) were among the most common types observed [16]. Within two weeks; our patient showed complete symptoms improvement and full resolution of systolic dysfunction. By reviewing the literature, most studies of TC cite echocardiographic and symptom improvement was occurring within days to weeks of diagnosis [17].

Conclusion

Inverted TC has been observed much less frequently in the literature compared to the classical apical ballooning syndrome. Our case can be added to the reported cases of stress-induced inverted TC. Knowledge about the perioperative upside-down TC lets the cardiologist quickly make this diagnosis, making timely recognition of this disorder crucial in initiating diagnostic and lifesaving treatment options. With supportive therapy for congestive heart failure early in this disease, the long-term prognosis will be excellent in the vast majority of patients.

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