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CASE REPORT

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A variant of Takotsubo syndrome concomitant with left atrial myxoma

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Abstract

We treated an 80-year-old Japanese woman who had Takotsubo syndrome (TTS) concomitant with a left atrial (LA) tumor. Left ventriculography revealed a variant of TTS. In cardiac surgery, the LA mass was successfully resected without embolism, with the pathological diagnosis of myxoma.

KEYWORDS

embolism, left atrial mass, myxoma, Takotsubo syndrome, variant type

1 | INTRODUCTION

Takotsubo syndrome (TTS) is a stress-related, reversible myocardial disorder.[1] Cardiac tumors are rare, observed in 0.002% of autopsies.[2] The combination of TTS and a cardiac tumor is thus extremely rare.[3,4] We treated a patient who had a combination of TTS and a cardiac tumor together with decompensated heart failure (HF). In a previous similar case, the tumor mass was removed, but some embolisms had occurred before the tumor's removal.[4]

2 | CASE REPORT

An 80-year-old Japanese woman was transported to our hospital because of persistent chest pain and dyspnea that had developed 4 h after she visited the grave of a relative. She reported having felt deep sorrow when she imagined her relative's death. Twenty years earlier, the patient had undergone surgery for breast cancer, and at that time, she was diagnosed with hypertension; antihypertensive medication was started. She had not received chemotherapy after surgery and was not pointed out for heart disease such as heart failure. On her arrival to our hospital, the patient's blood pressure was 100/75 mmHg, and her pulse rate was 120 beats per minute. She presented hypoxia requiring invasive positive-pressure ventilation.

Her initial electrocardiography showed ST elevation in leads V2–5 (Figure 1), and slightly increased levels of troponin T, creatine kinase, and creatine kinase MB were observed. Her chest radiography demonstrated pulmonary edema. Transthoracic echocardiography showed a movable left atrial (LA) mass (3.6 × 2.5 cm; Figure 2) and a depressed left ventricle (LV) ejection fraction (EF) at <45% with akinesis in the mid- to apical portion, with hyperkinesis in the basal portion of the LV. Transesophageal echocardiography revealed that the movable LA mass fit in the LV in conjunction with the heartbeat (Figure 3).

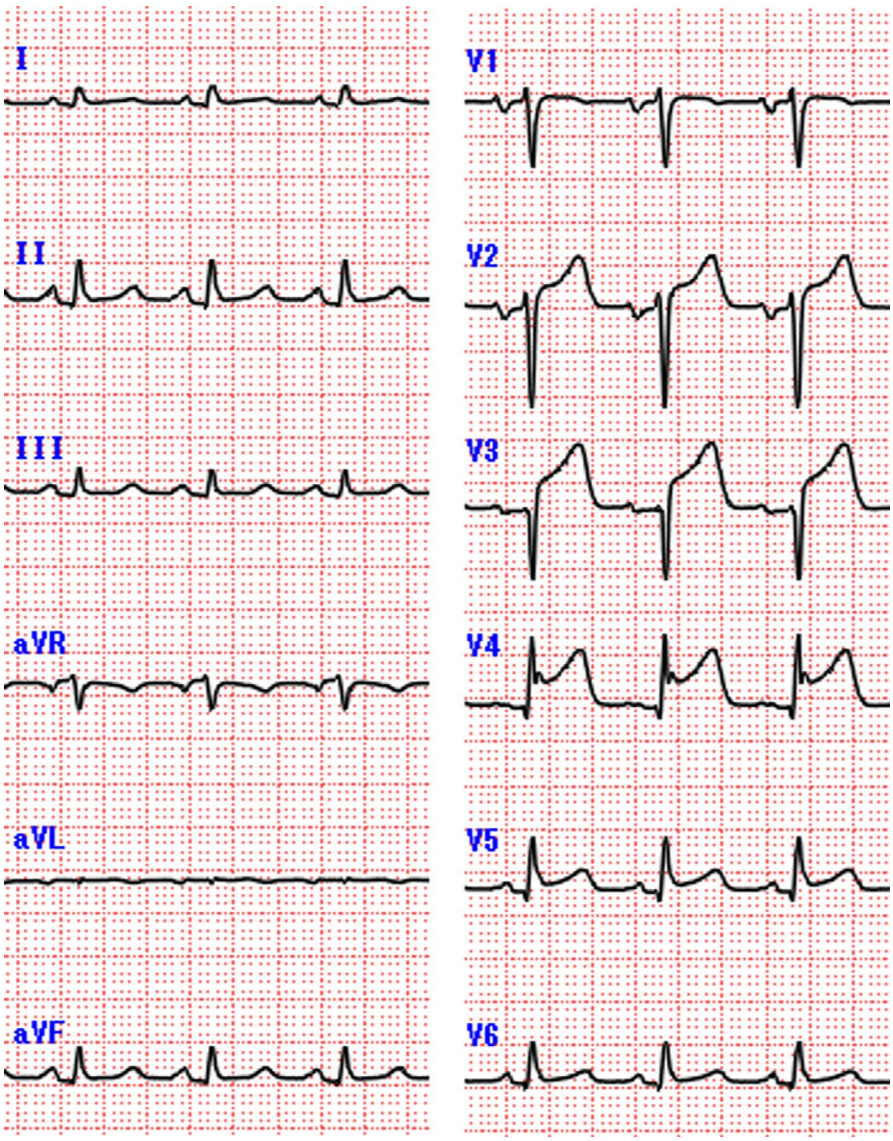

FIGURE 1 Electrocardiography on admission. In the initial electrocardiography, ST elevations were observed in leads V2–5

Coronary angiography showed no obstruction in the epicardial coronary artery (Figure 4A,B), and the feeding artery was derived from the right coronary artery (Figure 4A). Left ventriculography revealed akinesis in the mid- to large apex portion of the LV with vigorous contraction of the basal portion and the tip of apex segment (Figure 5A,B). The patient's clinical presentation was thus acute decompensated heart failure (HF) due to the large LA mass and a variant of TTS triggered by emotional distress. After the patient's hospitalization, we started medical treatment for HF, but her cardiovascular and respiratory status did not improve. Since the LA mass was large and had the potential to become lodged in the LV inflow tract, and since it was a movable mass, she was judged to be at a high risk of developing systemic embolism, and we decided to surgically remove the mass.

In the cardiac surgery, the 3.2 × 4.2 × 3.1-cm LA mass was resected. The pathology-based diagnosis was myxoma (Figure 6). On Day 3 after cardiac surgery, follow-up echocardiography showed that the LVEF had increased to 59%, and the contraction of previously akinetic regions was improved. The patient was discharged with no symptoms of dyspnea on Day 42 after the surgery. No systemic embolism occurred during her hospital stay.

3 | DISCUSSION

We encountered a patient with a variant of TTS concomitant with left atrial myxoma. The mass was successfully removed, without embolism. Although mass removal had been performed in a similar earlier case, embolisms had occurred before the surgery.[4] Generally, LV dysfunction in patients with TTS is transient and improves over the time. Early surgery might thus be recommended to avoid unstable circulatory dynamics; on the contrary,

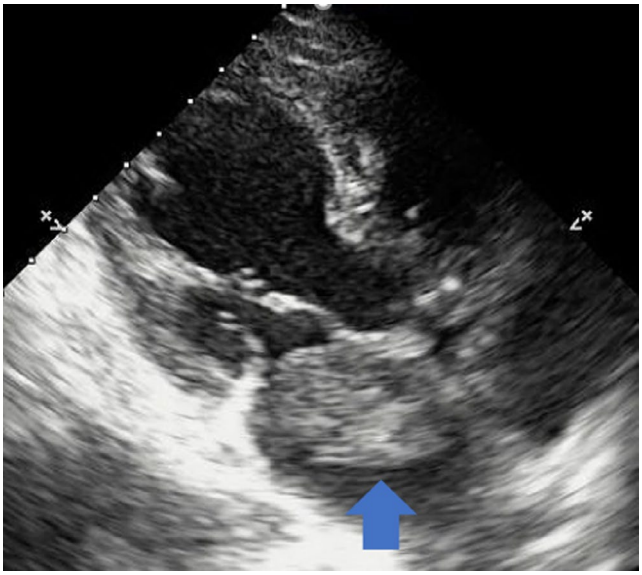

FIGURE 2 Transthoracic echocardiography revealing the nodular and mobile 3.6 × 2.5-cm mass in the left atrium (*blue arrow*)

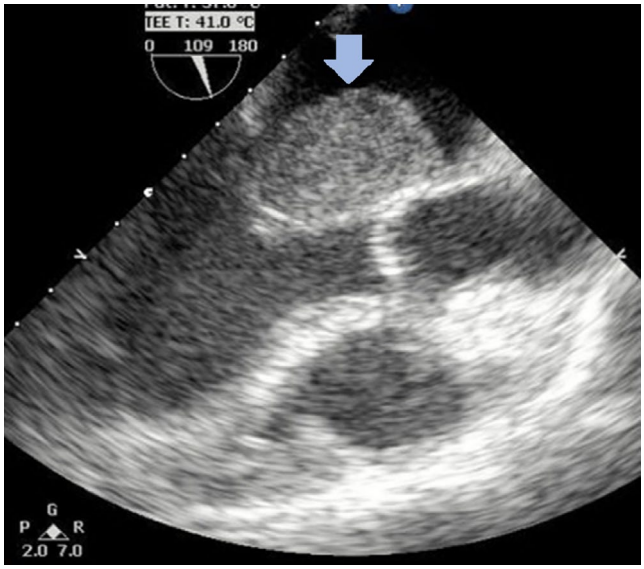

FIGURE 3 Trans-esophageal echocardiography depicting the mobile left atrium mass fitting in the left ventricle (*blue arrow*)

the development of an embolism from a cardiac tumor is unpredictable, and it is desirable to remove the tumor at an early point. In our patient, the hemodynamic status was not stable, but the LA mass was judged to be movable and she was at a high risk of experiencing an embolism. The LA mass was therefore removed at an early timepoint. As a result, the patient's hemodynamic status stabilized postoperatively and no embolic events occurred.

In our patient, akinesis in the mid- to large-apex portion with hyperkinesis in the basal portion and the tip of the apex segment was observed in the LV. This pattern of LV wall motion is similar to a mid-ventricular variant of TTS.[5] The most common type of TTS is the apical type (81.7%), followed by the mid-ventricular type (14.6%).[6] In the apical type of TTS, the impact of high levels of catecholamines up to the myocardium modulated by beta-adrenoceptor (βAR) gradients is suggested to contribute to a final common endpoint of acute apical dysfunction.[7] The βAR gradients in the LV might differ between the apical type and mid-ventricular type.

A cardiac myxoma can induce TTS, most likely through a variety of neurological mechanisms such as acute cerebrovascular embolism and the progressive involvement of the central autonomic network (CAN) through distant tumoral seeding.[8] It has been speculated that a cortical network comprised of the anterior cingulate cortex, amygdala, and insular cortex regulates the human cardiovascular system, and that the network of these cortical regions is necessary to regulate the CAN in response to emotional stress.[9,10] On the contrary, augmented systemic inflammation in patients with a cardiac myxoma is suggested to not only predispose patients to neurological complications but also to lower the threshold for TTS evolution in response to stressors.[8] In our patient, there have been no neurological complications associated with the myxoma. The emotional trigger related to her relative's death might

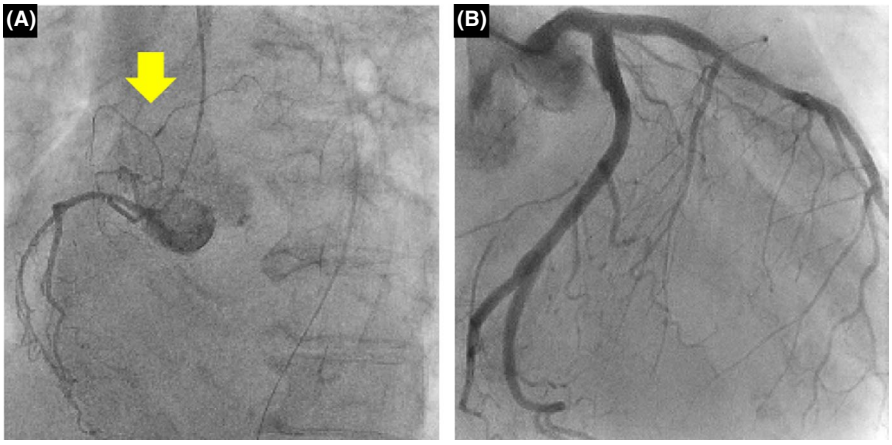

FIGURE 4 Coronary artery angiography. No stenosis was observed in the right coronary artery with the feeding artery derived (*yellow arrow*; A), left anterior descending artery, or circumflex artery (B)

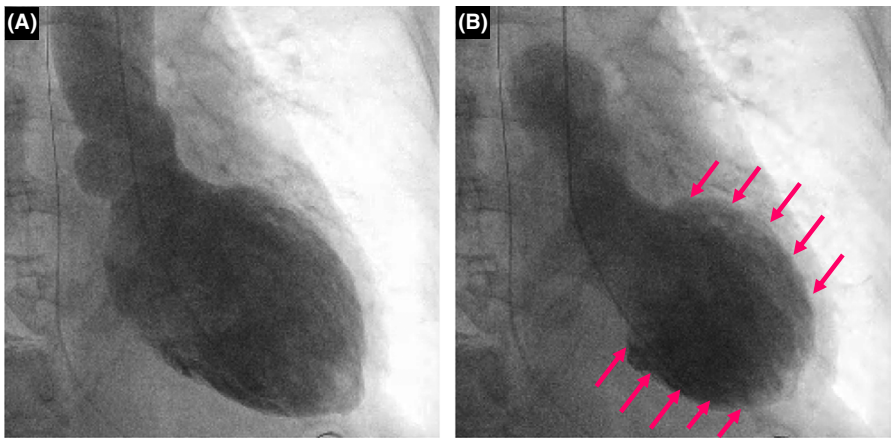

FIGURE 5 End-diastolic phase left ventriculogram (A) and end-systolic phase left ventriculogram (B). The extensive area around the mid- to large-apex portion showing akinesis (*arrows*), and the basal portion and the tip of the apex portion showing hypercontraction, especially in the end-systolic phase (B)

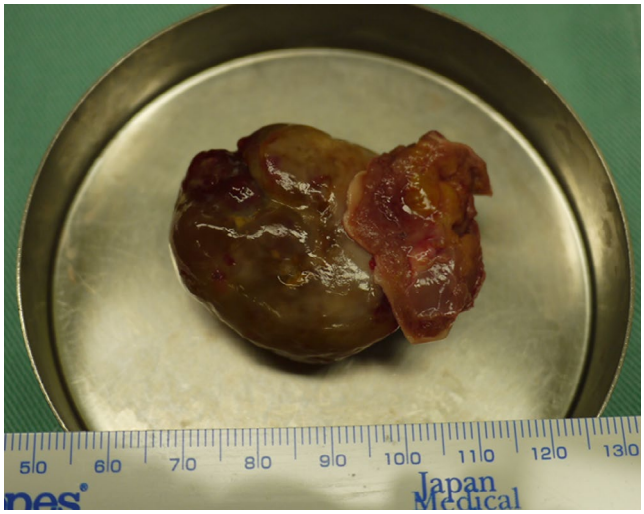

FIGURE 6 Gross specimen of the left atrial mass, observed as a hemorrhagic polypoid mass measuring 3.2 × 4.2 × 3.1 cm

have been associated with TTS due to a lowered threshold in relation to the increased inflammation caused by the patient's myxoma.

ACKNOWLEDGEMENT
Not applicable.

CONFLICT OF INTEREST
The authors declare that there is no conflict of interest.

AUTHOR CONTRIBUTIONS
NI, MN, and KD contributed to the conception of the case report. NI, MN, KD, MK, and NO participated in the analysis and interpretation of ultrasound. HT and AK participated in the analysis and interpretation of pathological examination. KD and MK assisted in the revision and supervised the overall production of this case report. All authors have read and approved the manuscript.

ETHICAL APPROVAL
No ethics approval required.

CONSENT
Informed consent was obtained from the patient.

DATA AVAILABILITY STATEMENT
The data that support the findings of this study are available from the corresponding author upon reasonable request.

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