An Unusual Case of Loffler Endomyocarditis after Takotsubo Cardiomyopathy Induced by Deep Neck Infection

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In this case, we herein have described a 72-year-old female patient with deep neck infection induced Takotsubo cardiomyopathy and idiopathic hypereosinophilic syndrome with Loffler endocarditis characterized by right atrial thrombus and right ventricular fibrothrombotic obliteration within two months.

Key Words: Cardiac thrombi • Hypereosinophilic syndrome • Takotsubo cardiomyopathy

INTRODUCTION

Takotsubo cardiomyopathy is a syndrome characterized by transient left ventricular apical or mid-apical ballooning and compensatory hyperkinesis of the basal segments without significant coronary artery stenosis. It is predominantly seen in postmenopausal women with emotional or physical stress and clinically mimics acute coronary syndrome.1 Idiopathic hypereosinophilic syndrome is described as eosinophilic infiltration of various organs, especially the heart, without the presence of eosinophilia-based reasons like parasitic infections, allergy or other known causes of eosinophilia.2,3 Cardiac involvement and thromboembolic complications are common causes of mortality and morbidity for patients with this chronic illness.4

Herein, we present a unique case report of a 72-year-old female patient with deep neck infection induced Takotsubo cardiomyopathy and idiopathic hypereosinophilic syndrome with Loffler endocarditis within two months.

CASE REPORT

A 72-year-old female patient with a history of deep neck infection caused by staphylococcus epidermidis and treated only with antimicrobial therapy (amoxicillin-clavulanic acid) within the previous two months was hospitalized because of the sudden onset of angina-like chest pain. Electrocardiography revealed ST-segment elevation in leads V1 through V3 (Figure 1). Her cardiac marker levels were elevated with a peak level of creatine kinase-MB isoform 37 ng/mL (normal reference range 0-20 U/L) and troponin-I 8.3 ng/mL (normal reference range 0-0.15 ng/mL). The patient underwent emergency cardiac catheterization, which disclosed no substantial epicardial coronary artery stenosis. Left ventriculography showed systolic ballooning of the apex and hypercontraction of the basal segment (Figure 2A-B), such that the patient was thought to have Takotsubo cardiomyopathy induced by deep neck infection. On her transthoracic echocardiography (TTE), the left ventricular apex was aneurysmatic, the ejection fraction (EF) was 40-45% and the right cardiac chambers were normal. The patient was thereafter discharged with a recommendation to undergo medical therapy. During her second month of outpatient care, clinical control TTE revealed endocardial thickening and fibrothrombotic obliteration of the right ventricle.
and a large immobile thrombus in the right atrium with normal ranged EF (Figure 2C-D). The left ventricular diastolic function was normal, and the right ventricular EF and tricuspid annular plane systolic excursion (TAPSE) were reduced. Review of color Doppler showed moderate tricuspid regurgitation. The patient was again hospitalized and low-molecular weight heparin therapy was started. Low extremity venous Doppler ultrasonography and pulmonary computerized tomography angiography had revealed no other evidence of thrombus. The patient also had 65% eosinophilia (11.500/μL) rate on her complete blood count. After exclusion of all other hypereosinophilia reasons, the patient was diagnosed with idiopathic hypereosinophilic syndrome with Loffler endocarditis and 60 mg per day methylprednisolone was started. On the 24th hour of the patient’s steroid therapy, her eosinophilia rate decreased to 0.2%. Warfarin was started with the intention of keeping an International Normalized Ratio of 2-3 on her follow-up. Three months later, follow up TTE showed that the right atrial thrombus and right ventricular infiltration had regressed completely so steroid therapy was terminated.

DISCUSSION

Takotsubo cardiomyopathy is a rare condition with a reported prevalence of 1-2% in patients presenting with chest pain. This condition is also known as the transient...
left ventricular apical ballooning syndrome, broken heart syndrome or stress-induced cardiomyopathy. It is predominantly seen in postmenopausal women and most commonly triggered by emotional stress or physical stress such as an unexpected death, infection, acute trauma and major surgeries.6

The clinical presentation of Takotsubo cardiomyopathy mimics acute coronary syndrome with chest pain, ST-T segment changes on electrocardiogram, mild elevation of serum cardiac enzymes, and transient left ventricular dysfunction with marked apical or mid-apical ballooning. The left ventricular dysfunction is usually reversible and normalizes within a few weeks or months. The exact cause and pathophysiology of Takotsubo cardiomyopathy is not well known, but several hypotheses include coronary vasospasm, abnormalities in coronary microvascular function, and catecholamine-mediated cardiotoxicity.1

Depending on the ventricular effected tissue area, it is named as typical and atypical. If the apical area is affected it is defined as typical, whereas an effect on the mid-ventricular area is atypical. While 60% of the patients with Takotsubo cardiomyopathy is designated as “typical,” the remaining 40% is therefore “atypical.” Both forms usually have excellent prognosis and a 7% recurrence risk, with a 1% mortality rate.5 There are no specific treatments for Takotsubo cardiomyopathy, but supportive measures such as administering oxygen and diuretics for pulmonary edema are needed in many cases.

Hypereosinophilic syndrome (HES) is characterized by persistent eosinophilia and eosinophil-mediated organ-system damage. HES has two subgroups: group A is primer HES which is also known as idiopathic HES, while group B is called seconder HES. Parasitic infections are the most common cause of seconder HES. Additionally, allergies, metastatic malignancies, endocrinopathies and leukemias are the other causes of secondary HES.4 Idiopathic hypereosinophilic syndrome is defined by the presence of a peripheral blood eosinophil count of 1,500/μL or greater for at least 6 months, exclusion of both secondary (including parasitic or viral infections, allergic diseases, drug-induced or chemical-induced eosinophilia, hypoadrenalism) and clonal eosinophilia, and evidence of organ involvement. If symptomatic and evidence of organ dysfunction is present, there is no need to wait for 6 months to start eosinophil-lowering therapy.7

Cardiac involvement is the most common extra-hematological manifestation of HES.2 Cardiac involvement and thromboembolic complications are common causes of mortality and morbidity. Endocardial fibrosis, mural thrombus and myocardial infiltration are responsible in cases of cardiac manifestation. Cardiac involvement with peripheral eosinophilia, irrespective of the reason, is called Loffler endocarditis.8 Heart failure, valve regurgitation, intracardiac thrombus formation, myocardial ischemia, arrhythmias, pericarditis, and syncope are the clinical manifestations of cardiac involvement.9 Although the left ventricle is more frequently affected, right or both ventricle involvement may still occur.1 However, atrial involvement is seldomly seen.

Eosinophils can survive in the tissues for weeks and the degranulation of activated eosinophils is thought to be responsible for the toxic damage caused to the heart. Toxins released by the eosinophils include eosinophil-derived neurotoxin, cationic protein, major basic protein and reactive oxygen species. These toxins may damage endothelial cells and myocytes, leading to necrosis and thrombosis and eventually culminating in endomyocardial fibrosis. Valvular involvement may manifest as regurgitation, and can be seen in both mitral and tricuspid valves secondary to intense inflammatory changes within the endocardium and papillary muscle dysfunction. In addition, there may be superimposed thrombus formation resulting in cardio-embolic manifestations.2

Echocardiography is the critical imaging modality for the diagnosis and follow-up of these patients. Although ideally, treatment should be directed to the cause of hypereosinophilia, corticosteroids may lead to regression of myocardial infiltrates when there is no etiological diagnosis.10 Response to corticosteroids can occur as early as five to six hours after initiation of treatment, which presents as a decreased eosinophil count. In steroid-resistant patients, cytotoxic drugs such as hydroxurea and interferon-A are alternative therapeutic approaches. Warfarin has been shown to be beneficial due to the association between Loeffler’s endocarditis and embolic events.

CONCLUSIONS

To our knowledge this is the first case report of
Takotsubo cardiomyopathy induced by deep neck infection in association with idiopathic HES with Loffler endocarditis in a patient.

**CONFLICT OF INTEREST, FUNDS, SUPPORT**

None.

**REFERENCES**