

SUCCESSFULLY TREATED ACUTE FULMINANT MYOCARDITIS INDUCED BY ULCERATIVE COLITIS WITH EXTRACORPOREAL LIFE SUPPORT AND INFLIXIMAB

HAN-KYUL KIM, MD¹, KUN IL KIM, MD, PHD², SUNG WON JUNG, MD, PHD³, HEE-SUN MUN, MD¹, JUNG RAE CHO, MD¹, NAMHO LEE, MD, PHD¹, AND MIN-KYUNG KANG, MD, PHD¹

DEPARTMENTS OF ¹CARDIOLOGY, ²CARDIOTHORACIC SURGERY, ³GASTROENTEROLOGY, KANGNAM SACRED HEART HOSPITAL, HALLYM UNIVERSITY MEDICAL CENTER, SEOUL, KOREA

We report a case of successfully treated acute fulminant myocarditis induced by ulcerative colitis with extracorporeal life support and infliximab. Myocarditis is a rare but crucial complication during an exacerbation of inflammatory bowel disease. In our case, we applied extracorporeal membrane oxygenation (ECMO) for cardiac rest under impression of acute myocarditis associated with ulcerative colitis, and added infliximab for uncontrolled inflammation by corticosteroid. As a result, our patient was completely recovered with successful weaning of ECMO.

KEY WORDS: Myocarditis · Ulcerative colitis · Extracorporeal membrane oxygenation.

INTRODUCTION

That is rare but crucial extra-intestinal complications during an exacerbation in patients with inflammatory bowel disease (IBD)—ulcerative colitis (UC) and Crohn's disease.^{1,2)} Cardiac complications occur particularly with UC,²⁾ and few cases reported patients with various manifestation from fulminant myocarditis to mild form.³⁻⁵⁾ Control of the underlying diseases is often associated with resolution of myocarditis and prevention of recurrence.⁶⁾ Here we report a young female patient with fulminant myocarditis induced by UC treated with extracorporeal life support and infliximab.

CASE

A 28-year-old woman was consulted for dyspnea and elevated cardiac biomarker (CK-MB: 11.4 ng/mL, troponin I: 0.512 ng/mL) from division of gastroenterology to cardiology. Sinus tachycardia and non-specific ST change was noted on her electrocardiography (ECG) and mild cardiomegaly and pulmonary congestion were noted on chest X-ray (CXR). She was suffering from exacerbation of UC treated with corticosteroid. Her blood pressure was 106/70 mm Hg and heart rate was

138 bpm. Initial echocardiography showed dilated left ventricular (LV) dimension and decreased LV ejection fraction (EF: 33%) with preserved LV tissue velocities (Fig. 1, Supplementary movie 1). We decided to apply extracorporeal membrane oxygenation (ECMO) for cardiac rest under impression of acute myocarditis because her coronary angiography showed normal coronary arteries (Fig. 2) in combination with global hypokinesia of LV. Follow up echocardiography performed the following day (Fig. 3, Supplementary movie 2) showed much depressed LV systolic function (EF: 10%) and myocardial tissue velocities (E': 10 → 3 cm/s, S': 6 → 2 cm/s). The 7th day after ECMO, massive hematochezia was occurred even with maintenance of corticosteroid (methylprednisolone 33 mg). Emergent sigmoidoscopy revealed diffuse maroon colored blood oozing from inflamed mucosa of transverse colon without discernable specific bleeding focus. Increased LV wall thickness (8 → 13 mm) and still depressed LV performance (EF: 16%, E': 2 cm/s, S': 3 cm/s) detected by echocardiography (Fig. 4, Supplementary movie 3), wherein we added infliximab (215 mg) infusion for uncontrolled inflammation. Two days later, removal of ECMO was possible because she sta-

• Received: October 20, 2015 • Revised: December 12, 2015 • Accepted: May 10, 2016

• Address for Correspondence: Min-Kyung Kang, Department of Cardiology, Kangnam Sacred Heart Hospital, Hallym University Medical Center, 1 Singil-ro, Yeongdeungpo-gu, Seoul 07441, Korea Tel: +82-2-2820-5294, Fax: +82-2-2846-4669, E-mail: homes78@naver.com

• This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

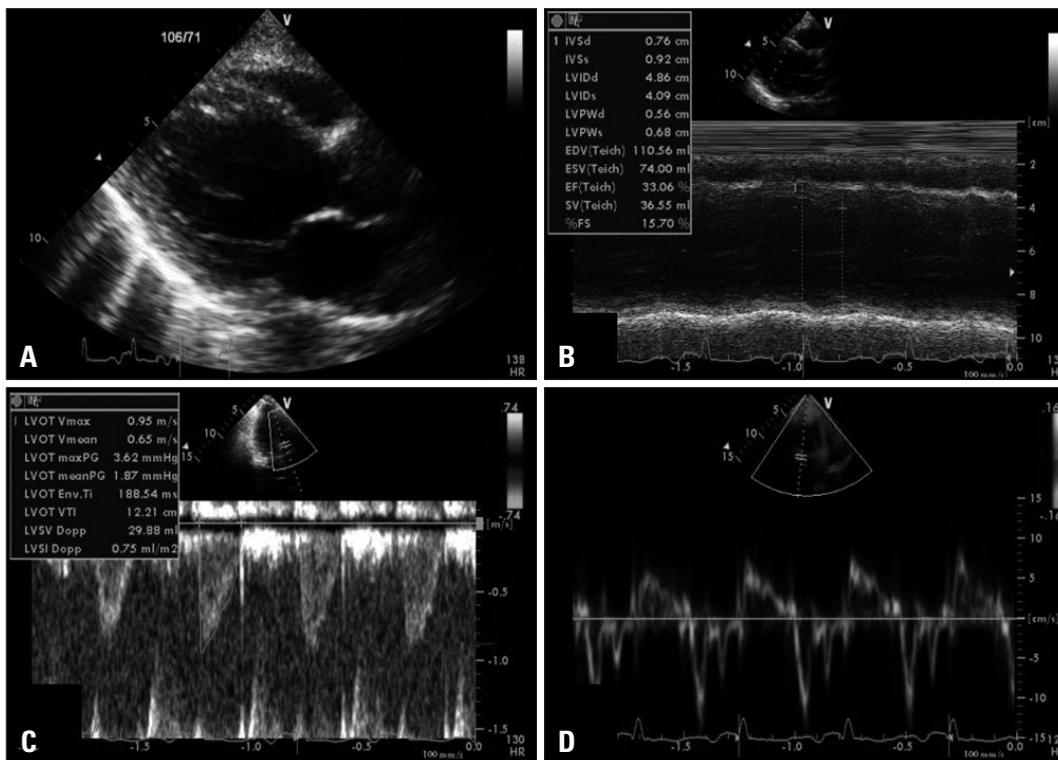


Fig. 1. Initial echocardiography shows dilated left ventricle by 2D (A) and decreased left ventricular (LV) ejection fraction about 33% by M-mode (B). Velocity time integral from LV out flow track showed 12 cm (C) but relatively preserved LV myocardial tissue velocities (D).

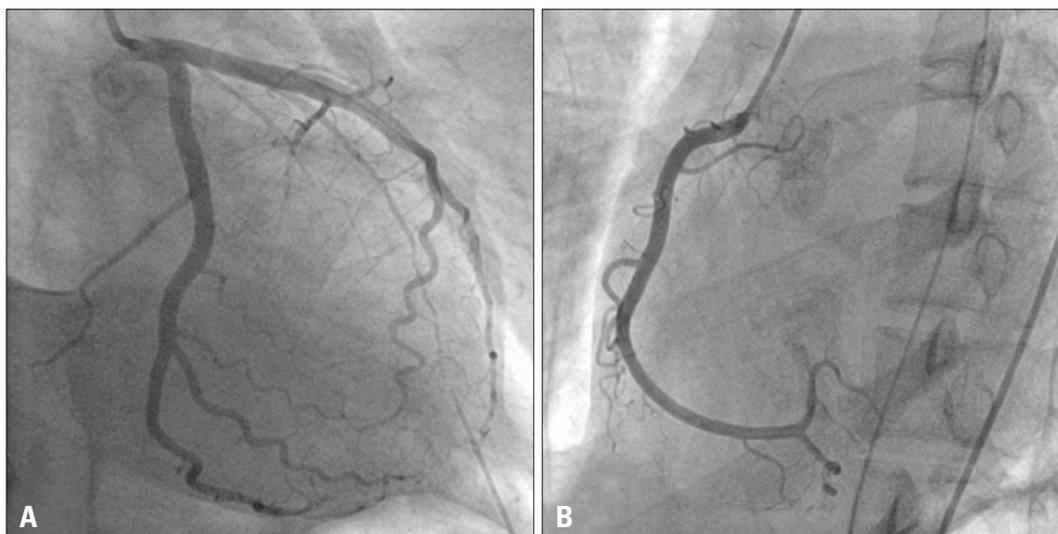


Fig. 2. Coronary angiography shows normal left (A) and right (B) coronary arteries.

bilized without further bleeding and echocardiography (Fig. 5, Supplementary movie 4) showed improved LV systolic function (EF: 16 → 60%) and myocardial tissue velocities (E': 3 → 5 cm/s, S': 2 → 7 cm/s). Last echocardiography (Fig. 6, Supplementary movie 5) showed normal LV wall thickness (11 → 8 mm), EF (65%), and myocardial tissue velocities (E': 12 cm/s, S': 10 cm/s). In addition, cardiac biomarkers (CK-MB: 11.4 → 8.4 → 5.8 → 1.4 ng/mL, troponin I: 0.512 → 0.298 → 0.099 → 0.025 ng/mL), ECG, and CXR were all normalized (Fig. 7). Now she is stable and followed regularly on outpatient basis

with maintenance of low dose steroid.

DISCUSSION

UC is known to be a severe disease with many extra-colonic manifestations, such as skin involvement, rheumatic problems, and ocular complications.²⁾ Here we present a young female patient with UC and fatal cardiac complication that were not resolved with corticosteroids.

In our patient, acute myocarditis was developed presenting dyspnea and elevated cardiac biomarkers during exacerbation of

UC in the maintenance of corticosteroid. We rapidly decided to apply ECMO for cardiac rest even her LV EF was not severely depressed and also with preserved LV myocardial tissue veloci-

ties. As a result, our decision was not so impetuous because echocardiography performed the very next day revealed severely depressed LV EF and myocardial tissue velocities. Unexpected

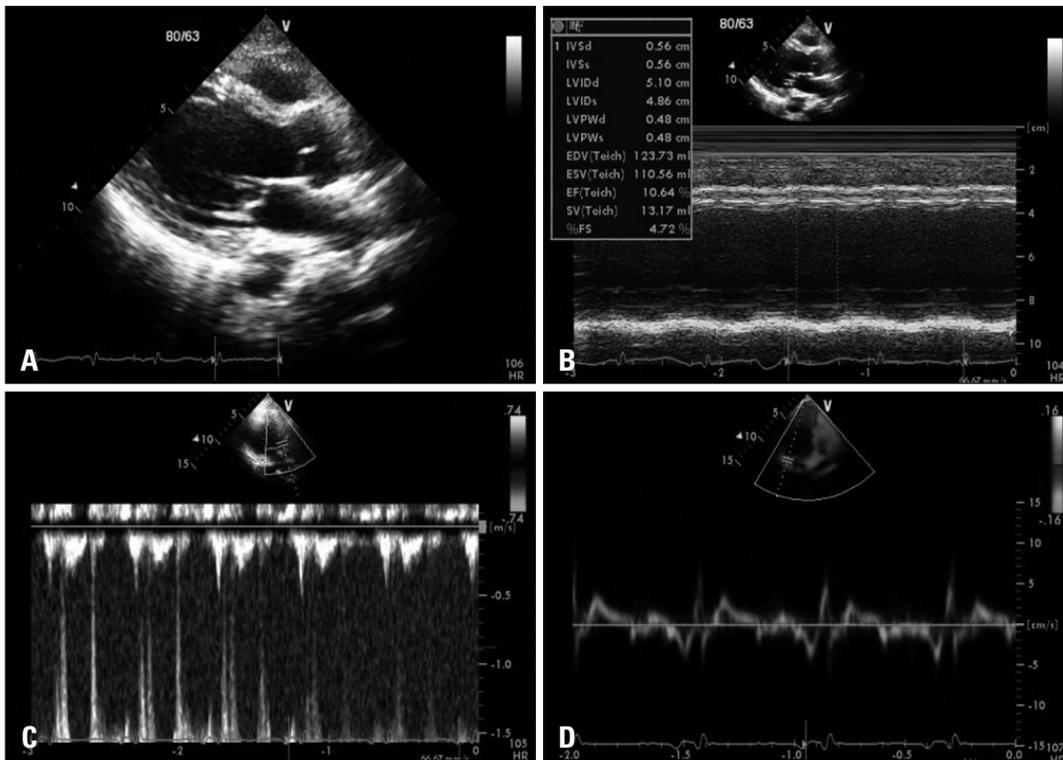


Fig. 3. Follow up echocardiography after extracorporeal life support shows further decreased left ventricular ejection fraction and velocity time integral (A, B, and C) and depressed myocardial tissue velocities (D).

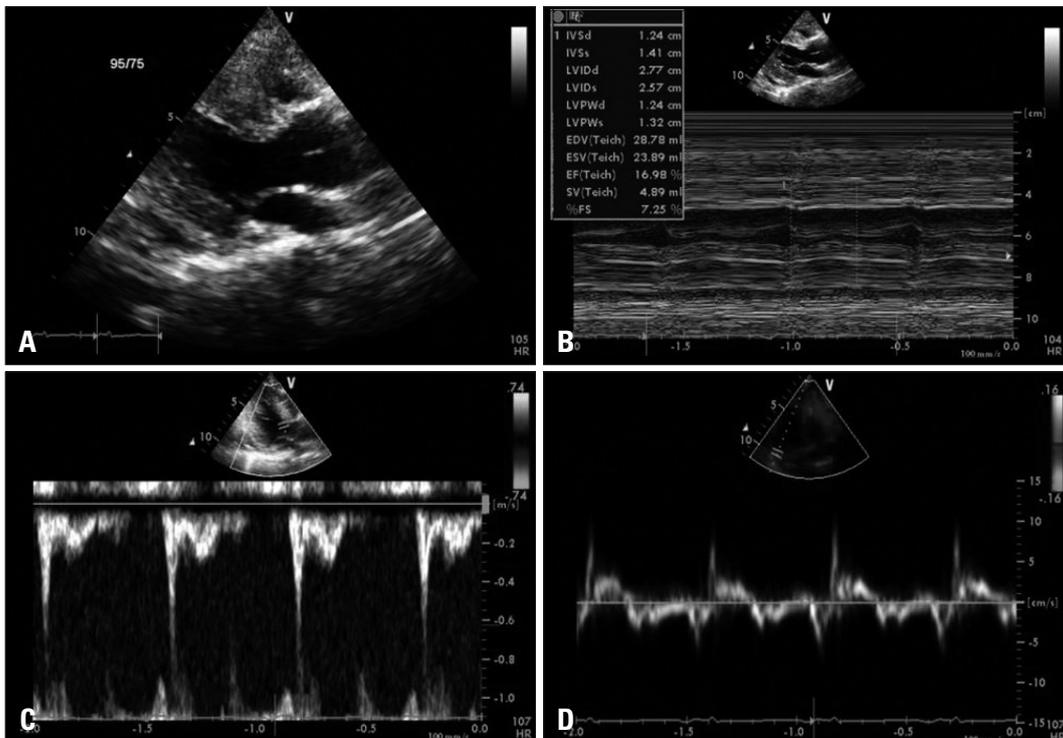


Fig. 4. Increased left ventricular wall thickness was detected after ECMO procedure (A and B) and still depressed velocity time integral from left ventricular out flow track and myocardial tissue velocities (C and D). ECMO: extracorporeal membrane oxygenation.

abrupt hematochezia was occurred despite of maintenance of corticosteroid (methylprednisolone 33 mg). Not only sigmoidoscopy showed uncontrolled inflammation of transverse colon, echocardiography also revealed increased thickness of myocar-

dium suggesting inflammatory phase of acute myocarditis.⁷⁾ Therefore we apply infliximab to control underlying bowel inflammation and acute management for UC. Follow up echocardiography after infusion infliximab, result showed improved LV

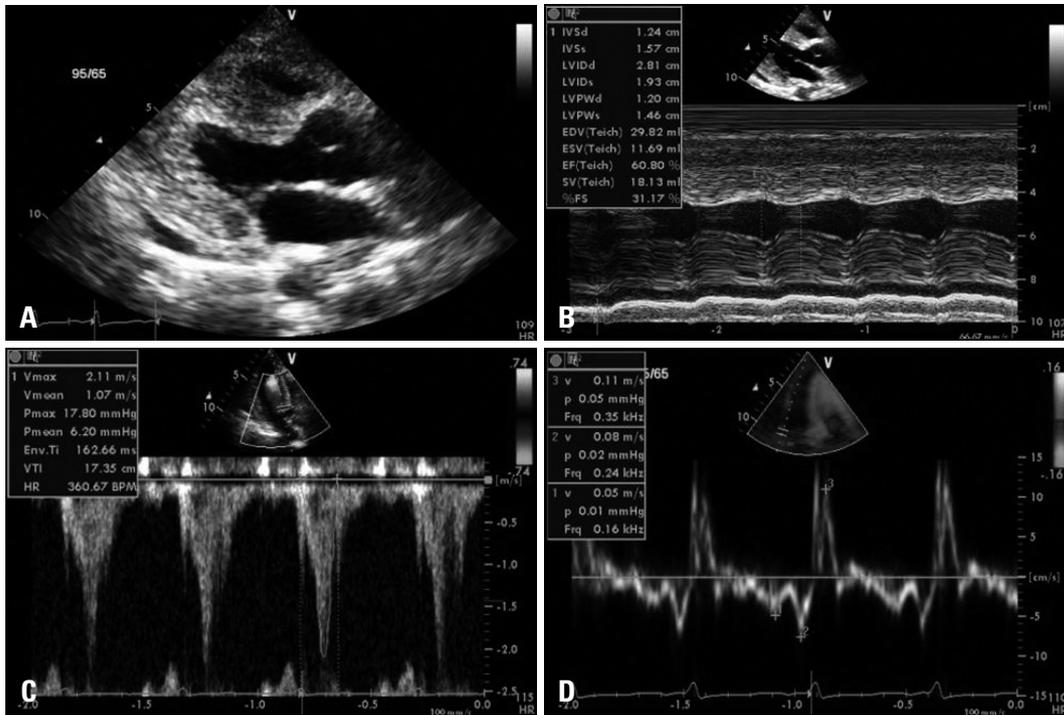


Fig. 5. The echocardiography after infusion of infliximab showed still increased left ventricular (LV) wall thickness but improved LV systolic function (A and B) and velocity time integral from LV out flow track and myocardial tissue velocities (C and D).

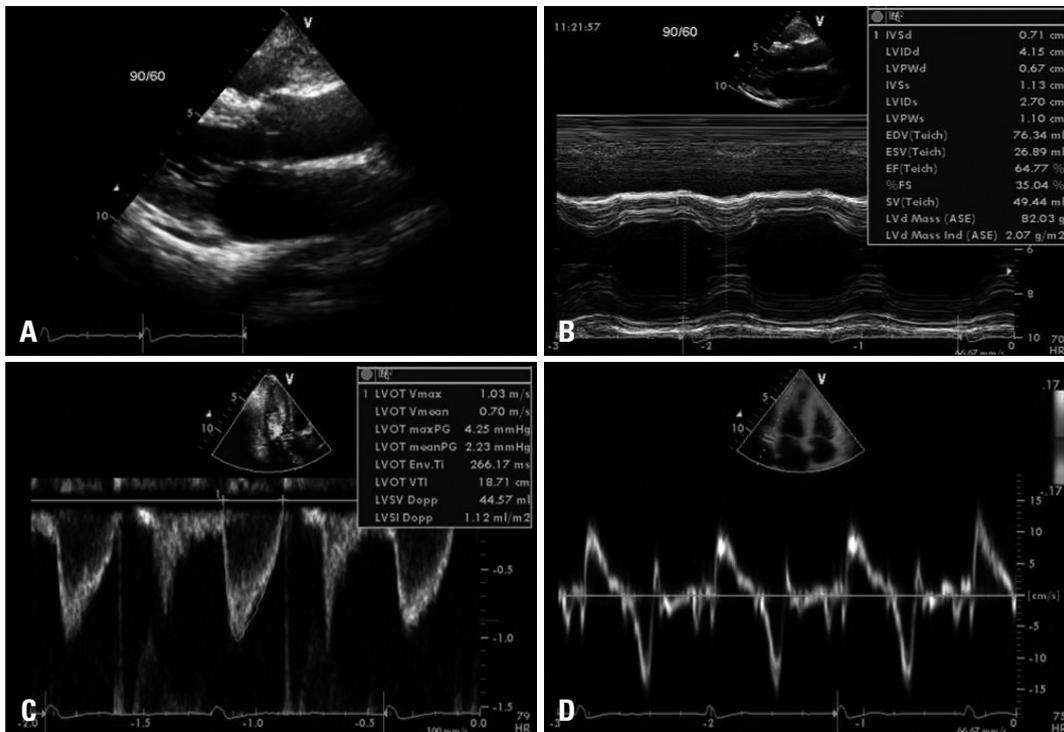


Fig. 6. The last echocardiographic study showed completely recovered myocardial function and resolved myocardial thickness. Normalized left ventricular (LV) wall thickness and improved LV systolic function (A and B) and normalized velocity time integral from LV out flow track and myocardial tissue velocities (C and D).

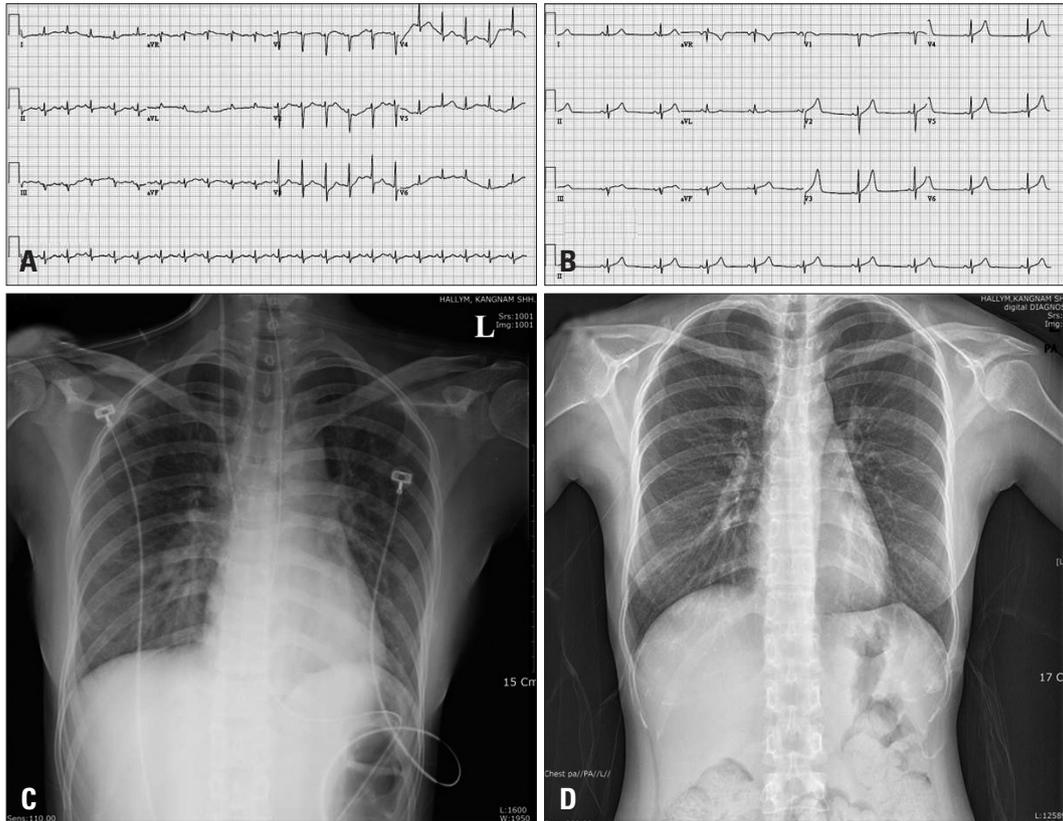


Fig. 7. Improvement of initial sinus tachycardia with non-specific ST changes on ECG (A) to normal sinus rhythm (B) and initial cardiomegaly and pulmonary congestion on chest X-ray (C) to normal (D). ECG: electrocardiography.

systolic function (EF: 16 \rightarrow 60%) and myocardial tissue velocities (E': 3 \rightarrow 5 cm/s, S': 2 \rightarrow 7 cm/s). Infliximab is a chimeric monoclonal antibody against tumor necrosis factor alpha used to treat autoimmune diseases. Although this agent has various adverse effects, some life-threatening, common to drugs in the class of immunosuppressants, patients with UC with treatment of infliximab was shown effective inflammation control.⁸⁾

In conclusion, we report a young female patient with fulminant myocarditis associated with UC successfully treated with ECMO and infliximab. Serial follow up of echocardiography and early decision of applying ECMO were particularly essential for management in our patient. Control of underlying disease might be helpful to improve cardiac function, in case of acute myocarditis due to underlying IBD.

SUPPLEMENTARY MOVIE LEGENDS

Movie 1. Initial echocardiography—dilated left ventricular (LV) dimension and decreased LV ejection fraction (ejection fraction: 33%) with preserved LV tissue velocities.

Movie 2. Second echocardiography—much depressed left ventricular systolic function (ejection fraction: 10%) and myocardial tissue velocities (E': 10 \rightarrow 3 cm/s, S': 6 \rightarrow 2 cm/s).

Movie 3. 7th echocardiography—increased left ventricular (LV) wall thickness (8 \rightarrow 13 mm) and still depressed LV performance (ejection fraction: 16%, E': 2 cm/s, S': 3 cm/s).

Movie 4. 9th echocardiography—improved left ventricular

systolic function (ejection fraction: 16 \rightarrow 60%) and myocardial tissue velocities (E': 3 \rightarrow 5 cm/s, S': 2 \rightarrow 7 cm/s).

Movie 5. Last echocardiography—normal left ventricular wall thickness (11 \rightarrow 8 mm), ejection fraction (65%), and myocardial tissue velocities (E': 12 cm/s, S': 10 cm/s).

REFERENCES

1. Mowat NA, Bennett PN, Finlayson JK, Brunt PW, Lancaster WM. *Myopericarditis complicating ulcerative colitis*. *Br Heart J* 1974;36:724-7.
2. Becker SA, Wishnitzer R, Botwin S, Eliraz A, Bass DD. *Myopericarditis associated with inflammatory bowel disease*. *J Clin Gastroenterol* 1981; 3:267-70.
3. Freeman HJ, Salh B. *Recurrent myopericarditis with extensive ulcerative colitis*. *Can J Cardiol* 2010;26:549-50.
4. Varnavas VC, Reinsch N, Perrey M, Nensa F, Schlosser T, Baba HA, Gerken G, Erbel R, Janosi RA, Katsounas A. *Recurrent lymphocytic myopericarditis in a young male with ulcerative colitis*. *Eur J Med Res* 2014;19:11.
5. Gruenhagen B, Alraies MC, Vakil KP, March SK. *Ulcerative colitis-induced myopericarditis*. *BMJ Case Rep* 2014 May 22 [Epub]. <http://dx.doi.org/10.1136/bcr-2014-204818>.
6. Kristensen KS, Høegholm A, Bohr L, Friis S. *Fatal myopericarditis associated with mesalazine*. *Lancet* 1990;335:605.
7. Kindermann I, Barth C, Mahfoud F, Ukena C, Lenski M, Yilmaz A, Klingel K, Kandolf R, Sechtem U, Cooper LT, Böhm M. *Update on myocarditis*. *J Am Coll Cardiol* 2012;59:779-92.
8. Iwasa R, Yamada A, Sono K, Furukawa R, Takeuchi K, Suzuki Y. *C-reactive protein level at 2 weeks following initiation of infliximab induction therapy predicts outcomes in patients with ulcerative colitis: a 3 year follow-up study*. *BMC Gastroenterol* 2015;15:103.