



Case Report

Cardiac rehabilitation in a patient with ankylosing spondylitis: A single-program, double-effect

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ABSTRACT

Ankylosing spondylitis (AS) is a chronic inflammatory disease which is characterized by the primary involvement of axial spine and sacroiliac joints and also extraarticular involvements including ophthalmological, cardiac, pulmonary, or neurological effects. Cardiac involvement in patients with AS has been reported to be 2 to 10%. Ventricular dysfunctions have been previously reported in patients with AS. However, there is only one case report describing primary cardiomyopathy due to AS and management with better control of inflammation and medications. Herein, we present a case of cardiomyopathy in AS in whom ejection fraction and New York Heart Association (NYHA) functional class improved and disease activity decreased after cardiac rehabilitation program

Keywords: Ankylosing spondylitis; cardiac rehabilitation; cardiomyopathy.

Ankylosing spondylitis (AS) is a chronic inflammatory disease which most often affects young men. Inflammation occurs in the sacroiliac joints, peripheral joints, spine, and entheses.^[1,2] Extra-articular manifestations such as uveitis, psoriasis, and chronic inflammatory bowel disease are frequently observed. Cardiovascular involvement is a rare, but considerable extra-articular manifestation, as cardiovascular disease risk is high among AS patients, compared to the general population and mortality from cardiovascular disease is about 20 to 40%.^[2,3] Myocardial involvement is not as well defined as involvement of the ascending aorta, aortic valves, and conduction system. If inflammation in AS is left untreated, it may lead to worsening of cardiac tissues with more involvement after longer periods of inflammation.^[4] Adversely, new onset development or progression of heart failure is a restriction for tumor necrosis factor antagonists for the treatment of patients with cardiac dysfunction due to AS.^[5,6]

Cases of cardiomyopathy have been reported in previously published echocardiographic studies.^[7-10]

Lui et al.^[4] reported four cases whose ejection fractions improved after better control of inflammation and medication. However, most researchers have tended to address into treating cardiomyopathy with medications rather than exercise therapy. Nevertheless, it is difficult to obtain a good management of cardiac involvement without exercise therapy, particularly when severe cardiac involvement together with high disease activity leading to contraindication for anti-rheumatic therapy occurs.

Herein, we present a case of cardiomyopathy in AS in whom ejection fraction and New York Heart Association (NYHA) functional class improved and disease activity decreased after cardiac rehabilitation program.

CASE REPORT

A 46-year-old male patient was admitted to our clinic with inflammatory back pain. He had inflammatory back pain, positive human leukocyte antigen (HLA)-B27 and pelvic radiograph showing

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Grade 4 sacroiliitis. He was diagnosed with axial spondyloarthritis. Initially, the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) score was 7, and the patient was given indomethacin 150 mg/day and sulfasalazine 2 g/day treatment. Despite the treatment with indomethacin for three months, the BASDAI scores did not improve and it remained above 4 points with increased C-reactive protein (CRP) values. Therefore, it was decided to start a biological agent; however, the patient had dyspnea and his functional capacity (FC) was Class III according to the NYHA. Before starting anti-tumor necrosis factor (anti-TNF) therapy, he was evaluated by a cardiologist due to his complaints of shortness of breath and chest pain during exercise. There were no abnormalities on his electrocardiogram and cardiac enzymes. As he had both hypertension and diabetes mellitus, he was under risk of atherosclerotic cardiovascular diseases. His serum lipid levels were normal. Coronary angiography was normal in search of ischemic heart failure etiology. He denied a history of rheumatic fever, excessive alcohol consumption, myocarditis which also could be attributed to ventricular dysfunction and dilatation. Decreased left ventricular systolic function, ejection fraction 25%, dilation of left cavities of heart, moderate mitral valve insufficiency, and normal right ventricular systolic function were reported on his echocardiogram.

He was given a diagnosis of non-ischemic cardiomyopathy by the cardiologist. Starting anti-TNF therapy was avoided, when he was diagnosed with cardiac failure. Therefore, indomethacin treatment was continued. The cardiac rehabilitation program was organized for the patient due to cardiac failure and AS in accordance with consensus document of the Heart Failure Association and the European Association for Cardiovascular Prevention and Rehabilitation.^[11]

An individualized aerobic exercise program was designed according to cycle ergometer test. Baseline results included a peak workload of 80 Watt at five min and 45 sec. The session was stopped by the patient, due to extreme fatigue. At the end of the test, the maximal O₂ consumption was 14.2 mL/kg/min. The metabolic equivalent (MET) value was 4.1 and maximal power per kg was 0.9. As the initial test values were very low, low intensity interval aerobic training was preferred. The cycle ergometer exercise program was started with warming with 10 Watts for four min and continued with 30 min of interval training including 120 sec of 60 Watts hard and 120 sec of 20 Watts for recovery, respectively. The program was completed

with cooling for four min with 10 watts. Before the main cycle ergometer program, the patient performed stretching exercises under supervision. The program was modified according to weekly assessments. After two weeks, strengthening exercises were added to main program. The strengthening exercises were designed according to 10 repetitions maximum and consisted large muscle groups of upper and lower extremity. Each muscle group was trained with 10 contractions of 80% of maximum load. The patient also received cardiac rehabilitation three times a week for eight weeks. At the end of eight weeks, the cycle ergometry exercises reached to warming with 25 Watts for four min and continued with 42 min of interval training including 120 sec of 100 Watts hard and 120 sec of 40 Watts for recovery, respectively. The program was completed with cooling for four min with 25 Watts.

Before treatment, six-minute walking distance was measured as 510 meters, BASDAI as 5.2, MET level as 4.1, and maximal loading as 80 watt. After the cardiac rehabilitation program, six-minute walking distance of the patient was calculated as 680 meters, MET level as 5.7, and maximal loading as 120 watts. During this period, pain and morning stiffness decreased significantly and BASDAI scores decreased to 1.3. The ejection fraction increased to 33% after an 11-week therapy. His FC regresses to Class I from Class III. The patient is still on an ongoing Phase-3 cardiac rehabilitation program with a low disease activity. Biological agent use became to be not necessary any more following cardiac rehabilitation. A verbal and written informed consent was obtained from the patient for the publication of this case report.

DISCUSSION

Ankylosing spondylitis is a systemic inflammatory rheumatic disease which is characterized by the primary involvement of axial spine and sacroiliac joints.^[12] In addition to musculoskeletal involvement, patients with AS also may show extra-skeletal involvements including ophthalmological, cardiac, pulmonary, or neurological effects.^[13,14]

Cardiac involvement in patients with AS has been reported to be 2 to 10%. While left ventricular dysfunction, aortitis, aortic regurgitation, pericarditis and cardiomegaly, involvement of conduction system and increased cardiovascular risk have been well-recognized in patients with AS, myocardial involvement still remains less well-defined.^[1,4,13,14] Cardiomyopathy which is a rare extra-articular manifestation of AS can be due to primary involvement of AS or secondary

to aortic and valvular involvement.^[14] In the present case, secondary causes of cardiomyopathy were excluded and cardiomyopathy was attributed to AS. It is assumed that underlying inflammation in AS also affects cardiac tissues resulting in cardiomyopathy.^[4]

Furthermore, the association between AS and ventricular dysfunction is still controversial. Deterioration of the ventricular functions in patients with AS has been reported in several previous echocardiographic reports.^[7-9,15,16] Lui et al.^[4] also reported four cases of primary cardiomyopathy due to no alternative explanation other than AS. However, Gould et al.^[8] compared patients with AS with age-, sex-, height-, and weight-matched healthy volunteers in terms of echocardiographic findings and found no statistically significant differences between them.

The present case and other reported cases by Lui et al.^[4] were found to be HLA-B27-positive. It was assumed that the heart and the joints were the major targets for HLA-B27 associated disease process.^[17] Interestingly, conduction disorders were found to be higher in HLA-B27-positive patients than in negative patients.^[18] However, Yildirim et al.^[15] found no difference in the systolic and diastolic function parameters between HLA-B27-positive and HLA-B27-negative patients with AS. The possibility and prevalence of HLA-B-27 positivity in patients with AS with cardiomyopathic involvement can also be a new area of future research.

Exercise therapy has been reported to be a corner stone of non-pharmacological therapy of AS in the 2010 update of the Assessment of Spondyloarthritis International Society/European League Against Rheumatism (ASAS/EULAR) recommendations for the management of AS. It has been stated that anti-TNF therapy should be initiated for patients with persistently high disease activity, despite conventional treatments according to the ASAS recommendations.^[3] Anti-TNF therapy is contraindicated and avoided in patients with moderate to severe heart failure NYHA Class III/IV.^[19] From this point of view, with the present case, we would like to emphasize that exercise therapy becomes the leading therapy in patients with high-disease activity, when medications are contraindicated and cardiac rehabilitation program including endurance exercises targets both disease remission and functional improvement in AS patients with heart failure.

On the other hand, it has not been fully clarified whether cardiac changes as a result of AS develop secondary to the primary involvement of cardiomyocytes or secondary to aortic and valvular

involvement.^[14] The present case report indicates the association between AS and cardiomyopathy and improvement of functional capacity and decreasing disease activity with cardiac rehabilitation for cardiomyopathy in patients with AS. To the best of our knowledge, the association with AS and cardiomyopathy can only be defined with cohort studies searching for the presence of tendency toward decreasing prevalence of cardiomyopathy with control of inflammation and exercise.

In conclusion, cardiac involvement in AS may be associated with increased disease activity and cardiac rehabilitation program may provide an improvement much beyond the known good effects of exercise in AS.

Declaration of conflicting interests

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