

CASE REPORT

Transient severe conduction disturbances associated with ankylosing spondylitis

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Email: shiro.hoshida@hosp-yao.osaka.jp**Abstract**

A 46-year-old man presented with advanced and complete atrioventricular block. He was diagnosed with human leukocyte antigen-B27-positive ankylosing spondylitis (AS) and treated with nonsteroidal anti-inflammatory drugs for AS. The severe atrioventricular block spontaneously improved and resolved after 3 months of therapy. Sequential cardiac magnetic resonance imaging demonstrated transient myocardial high-intensity signals in the basal septum close to the membranous portion of the septum. A pacemaker was not needed because of the reversible atrioventricular block.

KEYWORDS

ankylosing spondylitis, atrioventricular block, cardiac magnetic resonance, HLA-B27

1 | INTRODUCTION

Ankylosing spondylitis (AS) increases the risk of various cardiac manifestations, specifically conduction disturbances, aortic regurgitation, and coronary artery disease.¹ Although the prevalence of conduction disturbances ranges from 5% to 33%, complete atrioventricular block is quite rare.² There is a strong immunogenetic relationship between AS and human leukocyte antigen (HLA)-B27, which is linked to conduction disturbances and aortic regurgitation.³ Cardiac rhythm disturbances can lead to the implantation of a permanent pacemaker. Inflammation and fibrosis may play important roles in causing conduction disturbances. Furthermore, inflammation extending to the atrioventricular node, which is the membranous portion of the interventricular septum, may result in complete atrioventricular block.

Here, we report about a transient complete atrioventricular block accompanied with HLA-B27-positive AS. The transient cardiac tissue inflammation was identified by sequential cardiac magnetic resonance imaging.

2 | CASE REPORT

A 46-year-old man presented with general fatigue and dizziness on effort. Although he had chronic lumbago for 10 years, he never consulted an orthopedist. He was diagnosed with a severe conduction disturbance using an electrocardiogram (Figure 1A). His heart rhythm was 20-30 beats/minute with advanced and complete atrioventricular block and severe bradycardia. The serum levels of creatinine and electrolytes were within normal range and the plasma level of brain natriuretic peptide was 160 pg/mL on admission. A heart rate control therapy of a continuous isoproterenol injection was initiated on his admission day. His heart rhythm improved to Mobitz II second-degree atrioventricular block on the following day (Figure 1B), and his symptoms generally resolved. Transient advanced and complete atrioventricular blocks were observed only while he was sleeping. Ischemic heart disease was ruled out by a coronary artery angiogram, but mild coronary arteriosclerosis was observed. His carotid artery had mild arteriosclerotic changes based on ultrasonography. His cardiac function was normally preserved, with no signs of

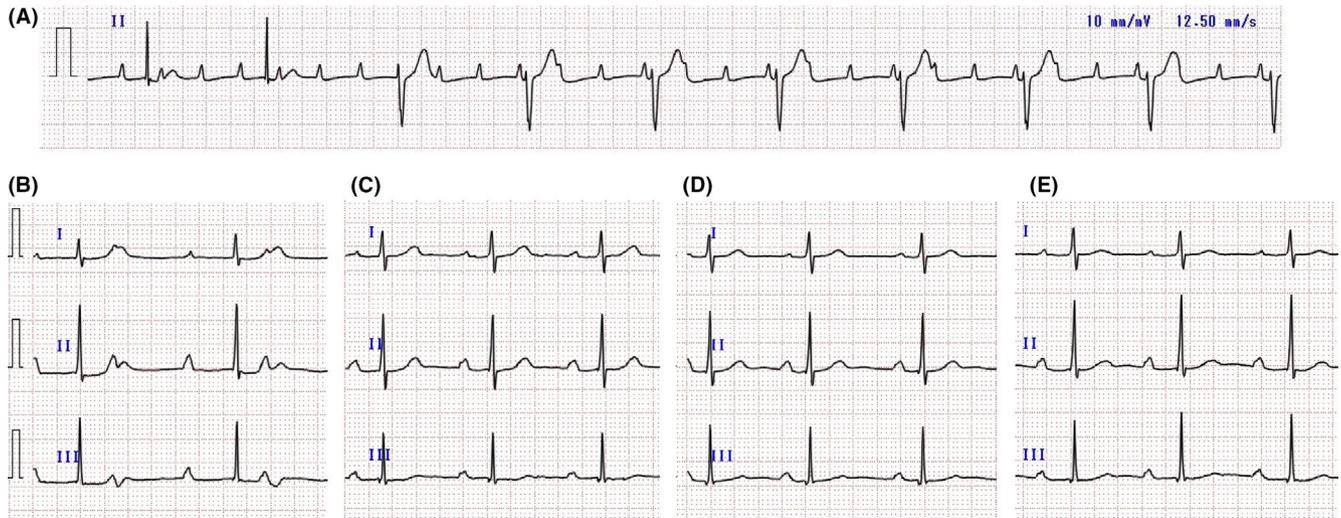


FIGURE 1 Electrocardiography showing advanced and complete atrioventricular block on the day of admission (A). The atrioventricular block spontaneously improved to a Mobitz II second-degree atrioventricular block next day (B) and to a first-degree atrioventricular block 2 wk after initiating celecoxib therapy (C). After 3 mo, the PR interval returned to normal limits (D). The recurrence of prolonged PR interval was observed 6 mo later with the upregulated inflammatory markers (E). The paper speed of the electrocardiogram was 12.5 mm/s only in Figure 1A. The paper speed of other figures (B-E) was usual (25 mm/s)

valvular diseases, including aortic regurgitation and basal septum degenerative changes, on echocardiography. He had a family history of AS; his father, younger paternal half-brother, and female cousin on the paternal side all had AS. X-ray and magnetic resonance imaging demonstrated bilateral sacroiliitis. He had clinical manifestations

of inflammatory back pain, limitation of lumbar motion, and limitation of chest expansion. The patient was finally diagnosed with AS. C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were slightly elevated at 0.66 mg/dL and 26 mm/h, respectively. Moreover, laboratory test results for HLA-B27 were positive.

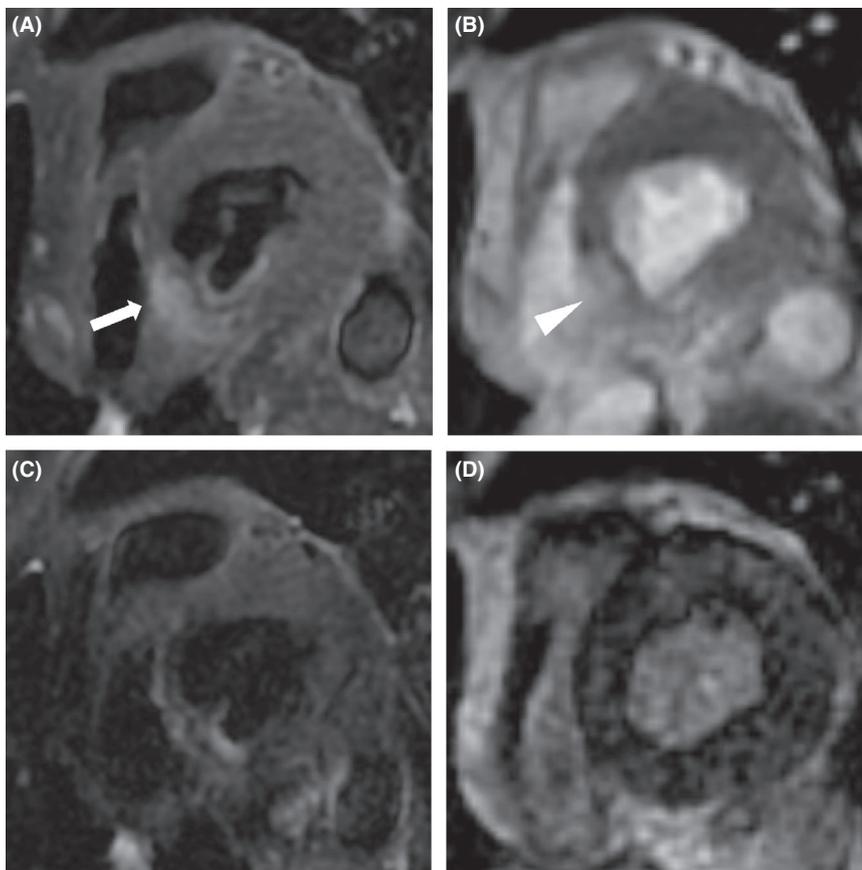


FIGURE 2 The patient was examined in the active phase by cardiac magnetic resonance imaging when he presented with the Mobitz II second-degree atrioventricular block. Fat-suppressed T2-weighted magnetic resonance imaging showed high-intensity signals in the basal septal myocardium close to the membranous septum (white arrow) (A). Late gadolinium enhancement indicated high-intensity signals in the same area (white arrowhead) (B). The high-intensity signals disappeared in the fat-suppressed T2-weighted (C) and late gadolinium enhancement imaging (D) at 5 mo after initiating therapy

Celecoxib (200 mg/day) was administered as a first-line medication therapy for AS after the definitive diagnosis. We changed the heart rate control therapy from isoproterenol to oral cilostazol (200 mg/day) because he did not consent to pacemaker therapy. He was eventually discharged, although the Mobitz II second-degree atrioventricular block persisted. Cardiac magnetic resonance imaging revealed a T2-weighted high-intensity signal (Figure 2A) and late gadolinium enhancement (Figure 2B) in the basal septum, suggesting inflammatory disease around the atrioventricular node and/or bundle of His. His conduction disturbance improved to a first-degree atrioventricular block after 14 days of celecoxib therapy (Figure 1C), and his symptoms completely resolved. The atrioventricular block was resolved 3 months later (Figure 1D). CRP and ESR decreased to 0.19 mg/dL and 11 mm/h, respectively, at that time. The severe conduction disturbance did not occur following medication therapy. The high-intensity signals completely disappeared in the cardiac magnetic resonance imaging scans after celecoxib therapy for 5 months (Figure 2C,D), showing the intensive, but reversible, inflammatory change of the septum in this case. However, the PR interval was prolonged again after celecoxib therapy for 6 months (Figure 1E), and CRP and ESR increased to 0.42 mg/dL and 15 mm/h, respectively.

3 | DISCUSSION

We report a case of AS diagnosed by the presence of a transient severe conduction disturbance. His severe atrioventricular block resumed within 2 weeks after initiating nonsteroidal anti-inflammatory drug therapy. Cardiac magnetic resonance imaging demonstrated transient enhancement in the myocardium close to the atrioventricular node and bundle of His. However, the patient did not require permanent pacemaker therapy. An intermittent complete atrioventricular block in patients with AS is quite rare, and focal inflammatory myocardium in the active phase is detected by cardiac magnetic resonance imaging. Furthermore, the PR intervals in the electrocardiogram had changed with systemic inflammatory markers.

Patients with AS have a twofold increased risk in second/third-degree atrioventricular block and pacemaker implantation compared with the general population.¹ Furthermore, male patients are more likely to have conduction disturbances. Dik et al. reported that the prevalence of first-degree atrioventricular block in patients with AS was 4.6% and that the PR interval was independently associated with disease duration,² suggesting that inflammatory burden affects the conduction system. However, the PR interval was not associated with inflammatory makers (i.e., CRP and ESR) in their cross-sectional study. The invasive electrophysiological studies showed that the level of atrioventricular block with HLA-B27-associated diseases was predominantly localized in the atrioventricular node.³ Conversely, the level of block in an acquired complete heart block is mostly below the bundle of His. Therefore, implantation of a pacemaker should be carefully considered in patients with HLA-B27-associated diseases, although pacemaker treatment is generally indispensable for cases who

present with high-grade atrioventricular block. This patient might need pacemaker implantation in the future.

Cardiovascular magnetic resonance is currently the gold standard for cardiac function and ventricular tissue characterization in patients with cardiovascular disease. Cardiovascular magnetic resonance can also be used to identify preclinical cardiac lesions in patients with connective tissue disease. Late gadolinium enhancement indicates cardiac tissue inflammation and fibrosis. However, late gadolinium enhancement itself does not always correlate with disease activity. Recently, T1 mapping has been used to calculate myocardial extracellular volume.⁴ Biesbroek et al. reported that the myocardial extracellular volume in patients with AS was correlated with the CRP concentration and ESR level.⁵ Novel cardiovascular magnetic resonance indices might provide disease activity of myocardial involvement in patients with AS. Further investigation is necessary to elucidate the relationship between myocardial extracellular volume and conduction disturbances.

CONFLICTS OF INTEREST

The authors declare no conflict of interests for this article.

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