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Case Report

Prolonged asystole during head-up tilt testing in a patient with Behcet's disease

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Abstract. Behcet's disease (BD) is a systemic inflammatory disorder and cardiac involvement is rare. Head-up tilt test is used for evaluating patients with neurocardiogenic syncope (NCS) and syncope of unknown etiology. A 31-year-old woman was examined for three syncopal episodes during the past two months, all of which preceded by nausea and sweating. Examinations including electrocardiography and echocardiography showed normal findings. A head-up tilt table testing was performed at an angle of 60 degrees. At about 8 minutes, syncope associated with bradycardia and asystole was observed. She became hypotensive, and there was a ventricular asystolic pause lasting 20 seconds, associated with loss of consciousness. She was placed in the supine position and cardiac massage was started. After 20 seconds, she slowly returned to sinus rhythm and regained consciousness. The patient was treated with dual-chamber (DDD-R) pacemaker implantation. During six months of follow-up, no major events occurred.

Key words: Behcet's disease, Head-up tilt test, vasovagal syncope, asystole

1. Introduction

Behcet's disease (BD) is a chronic inflammatory disorder with multisystemic manifestations. Cardiac involvement is rare; but pericarditis, myocarditis, valvular disease, arrhythmia, conduction impairment, and vasovagal syncope may occur (1,2).

Head-up tilt testing (HUT) is used as an in establishing for the diagnosis of neurocardiogenic syncope (NCS) and syncope of unknown etiology (3). The monitoring usually consist of heart rate and blood pressure measurements. Prolonged asystole is occasionally demonstrated during HUT and a subset of neutrally mediated syncope (4). The label 'malignant vasovagal syndrome' has been used some reports (4,5).

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The incidence of prolonged asystole during HUT is about 18% (>3 sec) and 9% (>5 sec) in patients with NCS (6). However, there are only a few reports describing asystole longer than 10 seconds (7,8). We demonstrate an adult female patient with frequent episodes of syncope in whom HUT was positive for a cardioinhibitory response with a prolonged asystole of 20 seconds.

2. Case report

A 31-year-old woman (married and has 2 children) presented to our dermatology outpatient clinic with complaints of recurrent oral aphtha and genital ulceration which had started 3 years previously. Considering the existing clinical findings and laboratory tests, a diagnosis of Behcet's disease was made and therapy was initiated with colchicine and antiinflammatory drugs. The patient, who had been diagnosed as having Behcet's disease and followed up for the previous 3 years. The patient was referred because of three syncopal episodes during the past two months, all of which preceded by nausea and sweating. The initial work-up including physical examination, chest X-ray, electrocardiogram (ECG), complete blood count, serum electrolytes, fasting blood sugar, thyroid

function studies, echocardiography, exercise stress test, neurological consultation, and Holter-ECG monitoring showed normal findings. She was not on any medication. Carotid sinus massage performed to exclude carotid sinus hypersensitivity was normal. A head-up tilt table testing was performed (3). The patient was subjected to an HUT at an angle of 60 degrees. At about 8 minutes of the test, syncope associated with bradycardia and asystole was showed without sublingual nitroglycerin stimulation. At the beginning of the test, his heart rate was 86/min. She became bradycardic and hypotensive (70/45 mmHg), and there was a ventricular asystolic pause lasting 20 seconds, associated with loss of consciousness (Figure). She recovered after being placed in the supine position and external cardiac massage was started. After about 20 seconds, she slowly returned to sinus rhythm and regained consciousness a few seconds later. The patient was treated with dual-chamber pacemaker (DDD-R) implantation. During six months of follow-up, no major events occurred and symptoms were controlled.

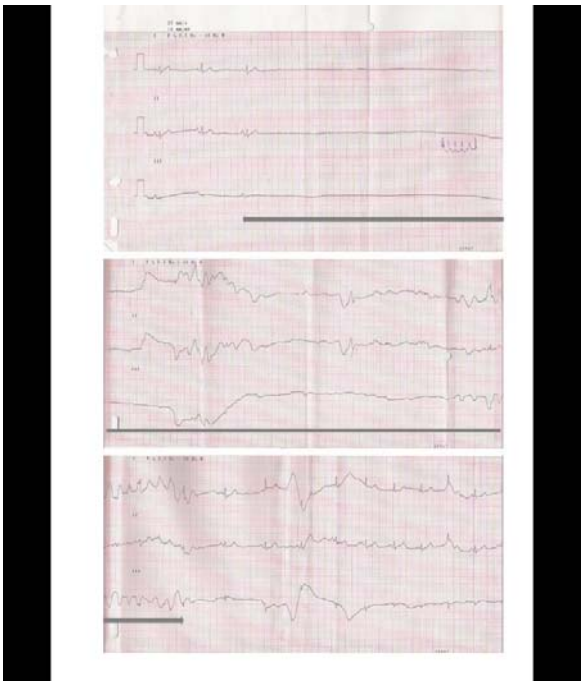

Fig. Electrocardiograms showing an asystolic pause lasting 20 seconds associated with loss of consciousness.

3. Discussion

We describe a case of an Behcet's disease female with frequent episodes of syncope, in whom HUT was positive for a cardioinhibitory response with a prolonged asystole of 20 seconds, which is not common in the literature (7,8). Although the incidence of prolonged asystole during HUT is about 18% (>3 sec) and 9% (>5 sec) in patients with NCS [6], this rate in BD is not known (6).

Vasovagal syncope is considered to have a good prognosis, although less is known about patients who are severely symptomatic, whose condition has been termed "malignant" (9). In a review of patients with asystole, it was demonstrated that *(first)* asystole during HUT did not necessarily imply a malignant outcome despite recurrences, *(second)* pacemaker or drug therapy did not influence outcome significantly, and *(third)* tilting protocol (angle) might influence time to and incidence of asystole during HUT (7). Milstein et al. suggested that life threatening cardiac asystole might occur in patients with the malignant form of NCS, and that this possibility should be considered when studying survivors of asystolic sudden cardiac arrest (4). In their study, all six survivors of suspected asystolic arrest with normal conventional baseline electrophysiological evaluation developed syncope during upright tilt provocation, with pauses of 16 and 20 seconds in two of them, respectively (4).

Head-up tilt testing is performed for the evaluation of NCS. The exact pathophysiological mechanism underlying HUT-induced asystole is not completely known. If prolonged asystole occurs during HUT, as seen in our case, external cardiac massage should be initiated without any delay to preserve irreversible ischemic damage (8). Although HUT is valuable in the assessment of syncope, it should be noted that it can yield false positive results especially in healthy young adults (10).

Tilt-induced prolonged asystole has been suggested to identify a distinct subgroup of patients with neurally mediated syncope, for whom management including permanent pacemaker implantation has been recommended. Several studies demonstrated improvement in the prevention of vasovagal syncope following pacemaker implantation (11,12).

In a randomized, controlled study, pacemakers were found to be superior to beta-blocker treatment in preventing syncopal recurrences (13). In our case, treatment with dual-chamber pacemaker (DDD-R) implantation resulted in improvement and during a follow-up of six months no major events occurred.

Although HUT is a very useful in diagnosis of BD patients with unexplained syncope, physicians should be aware of its potential complications such as prolonged asystole.

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