

Hiccups, a Rare Presentation of Infective Endocarditis: A Case Report

Hale Afshar, M.D.¹, Sina Bakhshaei, M.D.², Masoumeh Kahnooji, M.D.³, Mohsen Shafiepour, M.D.⁴,
Ahmad Alinaghi Langari, M.D.⁵

- 1- Assistant Professor of Pulmonary Diseases, Shahid Beheshti Hospital, Kashan University of Medical Sciences, Kashan, Iran
- 2- General practitioner, Clinical Research Development Unit, Afzalipour Hospital, Kerman University of Medical Sciences, Kerman, Iran
- 3- Assistant Professor of Cardiology, Cardiovascular Research Center, Institute of Basic and Clinical Physiology Sciences, Kerman University of Medical Sciences, Kerman, Iran
- 4- Assistant Professor of Pulmonary Diseases, Clinical Research Development Unit, Afzalipour Hospital, Kerman University of Medical Sciences, Kerman, Iran; Department of Internal Medicine, School of Medicine, Kerman University of Medical Sciences, Kerman, Iran (Corresponding author: Email: m.shafiepour@kmu.ac.ir)
- 5- General practitioner, Student Research Committee, School of Medicine, Kerman University of Medical Sciences, Kerman, Iran

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Abstract

Hiccup has a wide variety of etiologies irritating the hiccup reflex arc. It is usually benign and self-limited but can be an alarm sign for serious underlying conditions. Hiccup has been rarely reported in patients with infective endocarditis as a result of splenic involvement and phrenic nerve irritation. Herein, we report a 72-year-old man with the chief complaint of long-lasting hiccups (for a 1-month duration) who was diagnosed with infective endocarditis; however, his spleen was grossly intact. The bouts of hiccups repeated 2-3 times a day and lasted for 10-20 minutes. After the initiation of antibiotics, his symptoms waned for a while and appeared again, and then, the hiccups were eliminated. Long-lasting hiccups can be a presentation of serious medical conditions such as infective endocarditis, with or without splenic involvement. Hence, it is important to keep in mind these conditions to perform proper diagnostic and therapeutic workups.

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Introduction

Hiccup is defined as repetitive, involuntary myoclonic contractions of the diaphragm and intercostal muscles followed by sudden laryngeal closure, producing “hic” sound (1). Hiccup is a common experience, which is usually a self-limited condition, commonly induced by quick stomach distension and

irritation due to overeating or eating too fast. It can also occur due to aerophagia, ingesting spicy foods, immediate change in temperature of consumed foods, and anxiety. These conditions do not require treatment as long as they are not bothersome and subside spontaneously (2).

Persistent and intractable hiccups refer to the conditions in which hiccups last more than 48 hours and a month, respectively. The presence of intractable hiccups can be indicative of serious underlying pathologies, most of which are because of vagus and phrenic nerves irritation as a result of gastrointestinal (GI) disorders as well as mediastinal masses, goiter, diaphragm abnormalities, and malignancies (3). Other pathologic causes of hiccups include central nervous system (CNS) disorders (e.g. cerebrovascular accident, CNS tumors), metabolic disorders (e.g. hypokalemia, hypocalcemia, hypocarbia due to hyperventilation, uremia), psychogenic disorders, and drugs (4).

Infective endocarditis (IE) is a serious and uncommon clinical condition, defined as infection of the endocardial surface of the heart. IE has various systemic manifestations such as fever, unexplained stroke, and systemic septic embolism. It may rarely cause splenic abscess, which can irritate the diaphragm and lead to hiccups (5). Kalayci et al. reported a case presented with hiccups in a patient with pacemaker lead endocarditis. Patients with lead-related infection may present with nonspecific signs including fever, chills, and sweating. This study demonstrated a case of IE with vegetation on the tricuspid valve and pacemaker lead in a 66-year-old male patient with hiccup as an extremely rare symptom, who had underwent permanent pacemaker implantation three months ago (6). However, we report a case of infective endocarditis without evidence of splenic abscess that presented with hiccups. There was no more similar case in Iran or abroad for comparison.

Case Report

A 72-year-old man was admitted to the Shahid Bahonar hospital, Kerman because of fever, chills, and refractory hiccups for 1 month. His temperature was charted in the range of 38-40°C, which had no specific pattern. The bouts of hiccups repeated 2-3 times a day and lasted for 10-20 minutes. He was visited 3 times in the clinical office and received various antibiotics and some palliative treatments for hiccups. His symptoms waned for a while and appeared again. He also mentioned night sweats, loss of appetite, weight loss, weakness, and fatigue. There was no history of diarrhea, urinary symptoms, or cough. He had a history of hypertension, which was controlled with angiotensin-converting enzyme (ACE) inhibitors. He was nonsmoker and did not use drugs or alcohol.

On physical examination, the patient appeared pale and ill. His blood pressure was 130/80 mmHg; heart rate at 94 bpm; respiratory rate was 16 per minute; O₂ saturation was 91%, and the axillary temperature was 38°C. No lymphadenopathy was detected. Pulmonary sounds were normal and there was no crepitation. A grade 3/6 diastolic murmur was audible in the second right intercostal space. Examination of the abdomen and extremities were normal. There was no edema in both legs. The patient with these clinical manifestations was hospitalized with the diagnosis of unknown fever and hiccups for further investigation. Laboratory tests revealed a white cell count of 7800/μL, hemoglobin of 9.1 g/dl, platelet of 76000/μL, CRP of 95, ESR of 90, normal electrolytes, normal thyroid and liver function test, normal LDH, negative 3 blood cultures and urine culture. Tuberculin skin test was also negative. In the chest X-ray (CXR), no parenchymal lesion of the lung was detected (Figure 1). Abdominal sonography and CT scan were normal. Chest CT scan showed small bilateral non-loculated pleural

effusion, dilatation of ascending aorta and pulmonary trunk, cardiomegaly, prominence of both vascular patterns in lower zone of both lungs. Transthoracic echocardiography (TTE) revealed ejection fraction of 55-60%, pulmonary artery

pressure of 55 mmHg, mild mitral regurgitation, severe aortic insufficiency, and large mobile vegetation (1.26 × 0.57 cm) on the aortic valve (Figure 2).



Figure 1. Chest x-ray (CXR) showed no parenchymal lung lesion.

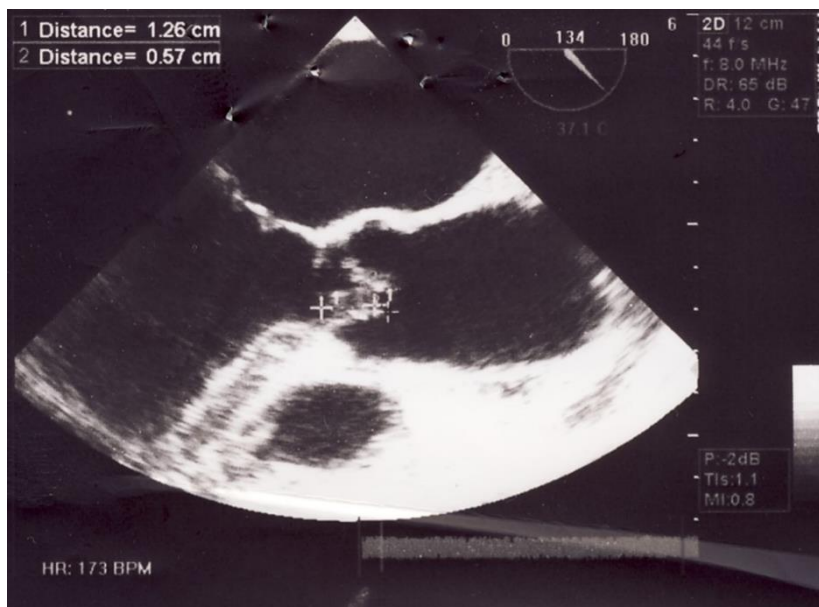


Figure 2. Transthoracic echocardiography (TTE) showed a large mobile vegetation (1.26 × 0.57 cm) on the aortic valve.

Intravenous antibiotics were prescribed promptly. Coronary angiography was performed and stenosis of two coronary arteries and multiple vegetation on the aortic valve were detected. He underwent combined aortic valve replacement and coronary artery bypass graft (CABG). The postoperative course was without any complications. The patient who was followed up 3 months later did not complain hiccups, he was in good condition and had no signs of infection, and all symptoms including fever and hiccups, disappeared after treatment.

Discussion

Clinicians refer to any febrile illness without an initially obvious cause as the fever of unknown origin (FUO). It has over 200 different causes based on many studies. The common causes are limited to several groups: infection, malignancy, noninfectious inflammatory disease, and miscellaneous (drugs, factitious, etc.) (7). After initial investigations, diagnostic clues determine which diagnostic procedures should be performed (8). Following this approach, we used transthoracic echocardiography to evaluate the diastolic murmur, and infective endocarditis (IE) was detected. IE has a wide variety of clinical features based on chronicity, including fever, chills, sweats, back pain, arthralgia, clubbing, neurologic and immunologic manifestation, etc. (7). Kalayci et al. reported a case presented with hiccups in a patient with pacemaker lead endocarditis (6). Celik et al. reported a case presented with hiccups in a patient with Twiddler's syndrome (9). As reported in a few literatures, IE can rarely present with intractable

hiccups. In all of these reports, concurrent splenic involvement (i.e. infarction or abscess) was suggested to be a potential cause of hiccups by irritating the diaphragm and phrenic nerve (5). However, in our case, no gross involvement of the spleen was observed, which makes IE a more probable independent cause of hiccups.

Hiccup reflex arc is comprised of the midbrain, C3-C5 of the spinal cord, phrenic nerve, vagus nerve, sympathetic trunk, and inspiratory muscles. Irritation of any part of this arc in the head, neck, chest or abdomen, will result in hiccups (10). Two potential afferent pathways of this arc originate from the heart. Cervical and thoracic segment branches of the vagus nerve are responsible for parasympathetic and sensory innervation of the heart. The heart sympathetic innervation is mediated by the cervical segment of the sympathetic trunk via celiac branches. Any process involving this complex may interfere with the reflex arc and result in hiccups. Hiccups can also be an unusual presentation of acute myocardial ischemia (10,11); however, the chronicity of ischemic heart disease in our patient cannot justify his recent symptoms (i.e. fever and hiccups).

Conclusion

Hiccup has a wide variety of etiologies. It is usually benign and self-limited but can be a presentation of serious underlying conditions such as infective endocarditis. Hence, it is important to keep in mind these conditions to perform the proper diagnostic and therapeutic workups.

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