CASE REPORT

Isolated Atrial Septal Defect Complicated by Tricuspid Valve Infective Endocarditis

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Abstract

Infective endocarditis (IE) associated with atrial septal defect (ASD) is extremely rare. However, tricuspid regurgitation (TR) secondary to right ventricular overload is a potential cause of IE, and once it occurs, the development of a paradoxical embolism may lead to fatal complications. We herein report the case of a 50-year-old woman who was admitted due to a persistent fever resistant to antibiotics. Echocardiography showed secundum ASD, moderate TR and a mobile vegetation measuring 15×10 mm attached to the tricuspid valve. Given the risk of developing a paradoxical embolism, urgent surgery was successfully performed.

Key words: congenital heart disease, atrial septal defect, infective endocarditis, tricuspid valve, embolism

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Introduction

Atrial septal defect (ASD) is a common congenital cardiac disease which accounts for one-third of all adult patients with congenital heart disease (1). Although an ASD causes a shunt in the flow between both atria, it does not predispose a patient to infective endocarditis (IE). Therefore, IE associated with ASD is extremely rare, and only a few previous reports exist in the available literature (2). We herein report an adult case of ASD complicated by tricuspid valve endocarditis in which tricuspid regurgitation (TR) secondary to right ventricular (RV) overload may have provided a substrate for IE.

Case Report

A 50-year-old woman with no past medical history except for untreated dental caries was referred to our hospital. In the primary care hospital, she was treated with 2 g/day cefazolin for the initial 2 weeks. Subsequently, cefazolin treatment was replaced with sulbactam/ampicillin (6 g/day) and administered for 2 weeks. However, the patient suffered from a persistent fever for one month. The physical examination showed a systolic murmur, a blood pressure of 110/60 mmHg, heart rate of 90 beats/min, a body temperature of 40°C and O2 saturation of 90% with ambient air. The electrocardiogram showed a sinus rhythm with an incomplete right bundle branch block and right axis deviation. The chest radiograph revealed a cardiac enlargement and infiltrate shadows in the lower lobes of both lungs. The laboratory findings included a white blood cell count of 10.5×10^9/L, C-reactive protein level of 15.0 mg/dL and B-type natriuretic peptide concentration of 166 pg/mL. In six blood culture examinations, one sample grew Gram-negative bacilli. Echocardiography showed a secundum ASD measuring 24 mm in diameter with a left-to-right shunting flow through the defect (Fig. 1A), as well as moderate TR (Fig. 1B) with RV and right atrial enlargement. The Qp/Qs ratio was calculated to be 2.9. No other concomitant disorders, such as a ventricular septum defect, aortic and mitral valve diseases or patent ductus arteriosus, were detected. The estimated systolic pulmonary artery pressure was 50 mmHg. We found a mobile vegetation measuring 15×10 mm attached to the anterior leaflet of the tricuspid valve (Fig. 1C, D). Notably, the vegetation had grown in size over the previous 3 days to 20×10 mm. Chest computed tomography (CT) showed a secundum ASD (Fig. 1E) and multiple nodules in the bilateral lung parenchyma.

We strongly suspected secundum ASD complicated by tri-
After ASD surgery, no IE was found before or after an 18-year observation period. Li et al. conducted a study on 5,000 adult congenital heart disease patients and observed a negligible risk for IE due to the slow velocity of the shunt flow. A slow flow in an ASD could reduce the risk of IE, but paradoxical embolism remains a concern. Indications for IE include tricuspid valve vegetations, which are often associated with isolated ASD. These findings were consistent with those of previous studies, which suggest that ASD is a potential cause of IE, although rare.

**Discussion**

We describe a case of tricuspid valve IE associated with an isolated ASD. Ventricular septal defects, patent ductus arteriosus, tetralogy of Fallot, and aortic valve abnormalities are congenital heart diseases recognized as risk substrates for IE. By contrast, an ASD has a negligible risk for IE due to the slow velocity of the shunt flow. In a population of 5,000 adult congenital heart disease patients, Li et al. did not find any IE before or after ASD surgery over an 18-year observation period. In the CONCOR National registry for adult congenital heart disease, Verheugt et al. determined the prevalence of IE in patients with ASD. In the CONCOR registry, nine ASD patients developed IE, although eight of these nine patients had concomitant lesions, such as a small ventricular septal defect. Therefore, IE associated with an isolated ASD is extremely rare. Although right-sided infective endocarditis (RSIE) occurs predominantly in intravenous drug users or in the presence of intracardiac electrodes or central venous catheters, the present patient did not have any of these risk factors. The patient had a history of dental caries, which could be a cause of transient bacteremia, with IE as the end result. The patient had teeth removed because of dental caries, and the antibiotic therapy was modified to a combination of ceftriaxone and gentamicin. Although brain magnetic resonance imaging (MRI) indicated no evidence of a stroke or intracranial aneurysm, given the risk of further pulmonary embolism and paradoxical embolism, urgent surgical treatment consisting of the removal of the vegetation and ASD patch-closure was successfully performed. Intraoperatively, we confirmed an isolated secundum ASD and found that the vegetation was attached to the anterior leaflet of the tricuspid valve. Microscopically, the vegetation consisted of aggregated leukocytes and fibrin. According to these findings, a diagnosis of tricuspid valve IE associated with isolated ASD was confirmed. The antibiotic therapy was continued for 4 weeks in the postoperative course. The patient’s recovery was uneventful and she was discharged 6 weeks after surgery.

**Figure 1.** TTE and TEE showing secundum ASD (A), moderate TR (B) and the vegetation attached to the tricuspid valve (arrows) (C, D: arrows). Computed tomography showing the enlargement of the RA and RV and the presence of ASD (E). ASD: atrial septal defect, LA: left atrium, RA: right atrium, RV: right ventricle, TEE: transesophageal echocardiography, TR: tricuspid regurgitation, TTE: transthoracic echocardiography, TV: tricuspid valve.
with ASD, unlike other RSIE, involves the risk of pulmonary and/or systemic embolism. RSIE with ASD has an anatomical feature distinguishing it from other RSIE, which is characterized as an open communication between both atria; the interatrial communications are bidirectional, even in the absence of pulmonary vascular disease and severe pulmonary hypertension. Theoretically, although left-right shunting is dominant in ASD, right-left shunting can be provoked by coughing or the Valsalva maneuver, which elevates the right atrial pressure. Moreover, atrial flutter and fibrillation, which alter the interatrial pressure balance, are observed in 21% of adults older than 40 years of age with an increasing frequency over time (5). Indeed, Bannan et al. reported that the incidence of adults with ASD who presented with paradoxical embolism was higher than expected at 14% (6). RSIE accounts for 5-10% of all IE cases, and most RSIE cases can be treated medically, although surgery may be necessary in some cases, including uncontrolled septicemia, right-sided heart failure, recurrent episodes of pulmonary embolism, and vegetations >20 mm in diameter (7). Given the risk of paradoxical embolism, however, tricuspid IE with ASD may require early surgical treatment to avoid fatal complications. Although the preoperative MRI and CT scan indicated no evidence of a stroke or other systemic infections in the present patient, urgent surgery was successfully performed and she had an uneventful postoperative course.

The authors state that they have no Conflict of Interest (COI).

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