

Endocarditis and bacterial brain abscess in a young woman with a single atrium, patent ductus arteriosus, and Eisenmenger syndrome

A case report

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Abstract

Rationale: A single atrium is a rare congenital heart disease (CHD) involving zero atrial septal traces and preserved intact ventricular septum and atrioventricular valves, requiring careful surgical intervention. However, developing to Eisenmenger syndrome (ES) makes the surgery complicated. Based on bidirectional cardiac shunting, vegetation easily develops in case of bacterial infection.

Patient concern and diagnoses: We reported a 35-year-old woman with a single atrium, patent ductus arteriosus, pulmonary hypertension, and ES who developed infective endocarditis on her left ventricular outflow tract and complicated cerebral abscess and who underwent challenged medical treatment.

Intervention: Infection was successfully controlled after 4-time change in antibiotics over 4 months. However, surgery is complicated for her.

Outcomes: The patient presented a relatively good outcome during follow-up for >6 months.

Lessons: This case report suggests that patients with complex CHD should accept surgery therapy earlier before developing ES. It is imperative to avoid invasive interventions to prevent infectious endocarditis.

Abbreviations: CHD = congenital heart disease, ECD = endocardial cushion defect, ES = Eisenmenger syndrome, PDA = patent ductus arteriosus.

Keywords: cerebral abscess, congenital heart disease, endocarditis, single atrium

1. Introduction

Complete endocardial cushion defect (ECD) involves a large primum atrial septal defect and an inlet ventricular septal defect of variable size. Complete ECD accounts for approximately 3% of congenital heart defects.^[1] For patients with a large atrial

septal defect, only one large heart valve (common valve) is commonly observed instead of two distinct atrioventricular valves (mitral and tricuspid). Surgery should be performed after birth depending upon severity. When pressure in pulmonary circulation exceeds systemic pressure, bidirectional intracardiac shunting occurs (Eisenmenger syndrome [ES]), causing difficulty in treatment. Patients gradually present with breathlessness, fatigue, chest pain, and syncope.^[2] Based on special hemodynamics, vegetation easily develops in case of bacterial infection. Here, we report a young woman born with a single atrium and who developed endocarditis on the left ventricular outflow tract and complicated cerebral abscess.

2. Case report

A 35-year-old woman was born with a single atrium and patent ductus arteriosus (PDA) and developed severe exertional dyspnea. She refused surgery or medical therapy due to economic problems. She was admitted with chills, fever (highest 39.5°C), cough, and headache for six days. Hypotension and hypoxia were presented on admission. Echocardiogram showed PDA (right-to-left shunting) and partial atrioventricular defect with a large atrial septal defect in the remaining two distinct atrioventricular valves, resulting in a single atrium (Fig. 1A–C). An oscillating intracardiac vegetation (1.3 × 0.9 cm) attached on the left ventricular outflow tract was detected near the anterior mitral leaflet root with an increasing speed of 2.57 m/s blood (Fig. 1D). Color flow imaging and pulsed wave Doppler demonstrated bidirectional cardiac shunting with dominant left-to-right shunting. The estimated pulmonary arterial systolic

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Informed written consent was obtained from the patient for publication of this case report and accompanying images.

The authors have no conflicts of interest to disclose.

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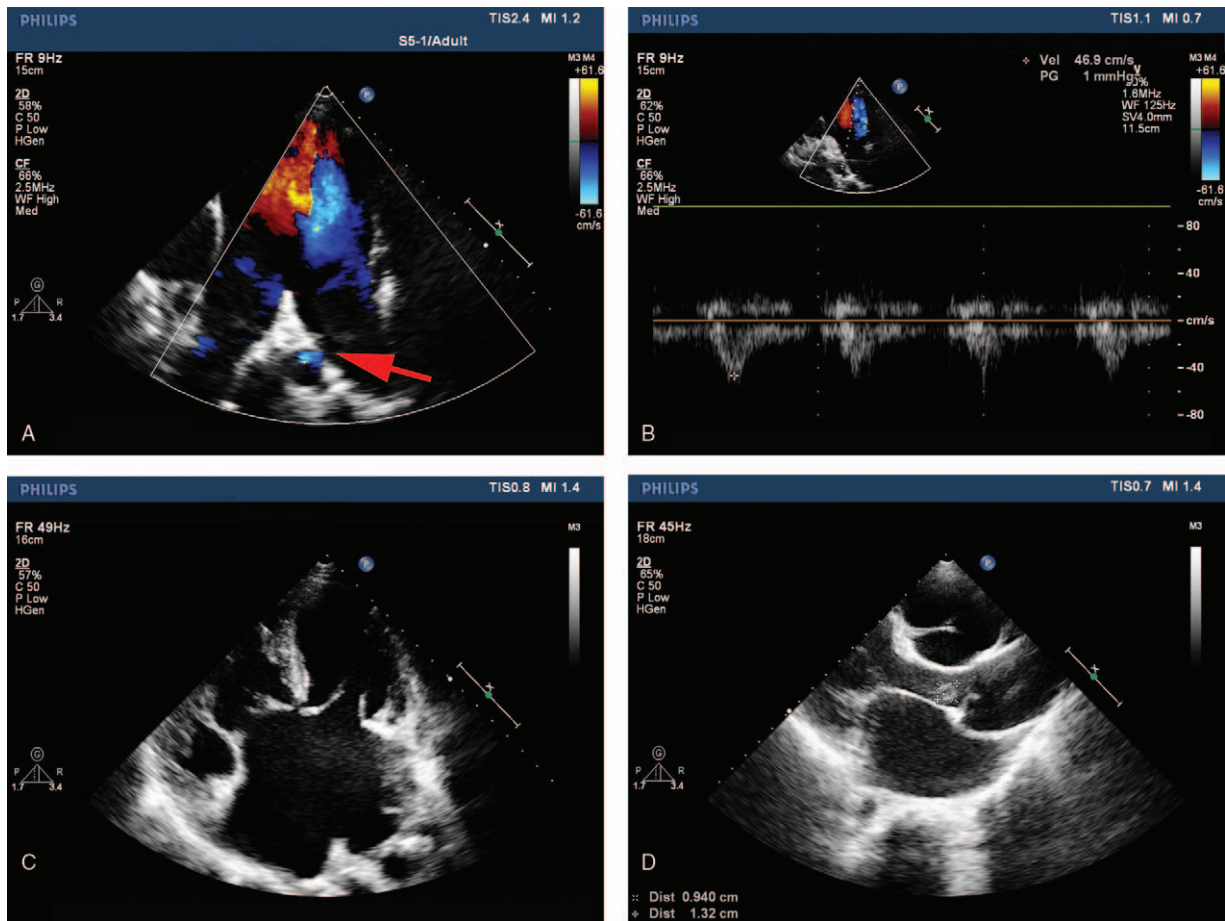


Figure 1. Transthoracic echocardiogram. (A) Transthoracic parasternal short-axis view of pulmonary artery with two branches, demonstrating enlarged pulmonary artery with PDA (red arrow) observed on color Doppler. (B) Pulsed wave Doppler showed right-to-left shunting from the left pulmonary artery toward the descending aorta. (C) Transthoracic apical four-chamber view showing a partial atrioventricular defect with a large atrial septal defect in remaining two distinct atrioventricular valves (mitral and tricuspid). (D) Transthoracic parasternal long-axis view of left ventricular outflow tract demonstrating an oscillating intracardiac vegetation near the anterior mitral leaflet root.

pressure was 102 mmHg. ES and pulmonary hypertension were diagnosed. Head magnetic resonance imaging (MRI) showed a ring-enhanced lesion (2.0 × 1.2 cm), which was surrounded by a large range of edema, in the left temporal brain, suggestive of bacterial cerebral abscess (Fig. 2A–D). However, blood cultures obtained from separate venipuncture sites with the first and last samples drawn 3 h apart were negative, as were cerebral spinal fluid cultures.

Infectious endocarditis was diagnosed based on one major criterion (vegetation on echo) and three minor criteria (predisposing heart condition, temperature >38°C, and cerebral abscess with elevated infectious indexes).^[3] According to the medical history, the infection source might be the site of injection of painkiller for dysmenorrhea. Empirically, being suspicious of *Dermatococcus*, the patient was administered with high-dose penicillin G (high blood brain barrier permeability). However, antibiotics were changed to vancomycin because of severe allergic reaction.^[3] During treatment, symptoms were relieved, and decreasing abscess lesion was observed on head MRI, showing evidence of effective antibiotics that covered Gram-positive bacilli. Until severely low leucocyte counts were observed, linezolid was considered an alternative to vancomycin. Given the recurrent decreased leucocyte, ceftriaxone was selected

as the last de-escalation of antibiotics for its high blood brain barrier permeability^[4] and low cross-activity against penicillin (different side chains from penicillin).^[5] After the 4-month treatment, repeated MRI showed significantly faded abscess lesion (Fig. 3A–D). However, repeated echocardiogram showed no change in vegetation. Apropos of vegetation removal, such complicated heart conditions and ES result in high-risk surgery. The patient was discharged with continuous pulmonary vasodilators and oxygen supplement, achieving a relatively good outcome during follow-up for >6 months. Her response to therapy and absence of recurrent embolic events indicated complex of surgery.

3. Discussion

Positive blood cultures are a major diagnostic criterion in endocarditis and key to identifying pathogenic agents.^[3] However, in this case, recurrent blood cultures were all negative. We realize that the intensity of bacteremia may be negligible; however, an investigation detected <50 colony-forming units per 1 ml blood in the majority of patients.^[3] We considered the following possible reasons for the negative blood cultures. Firstly, microbiological techniques were inadequate, and infection with

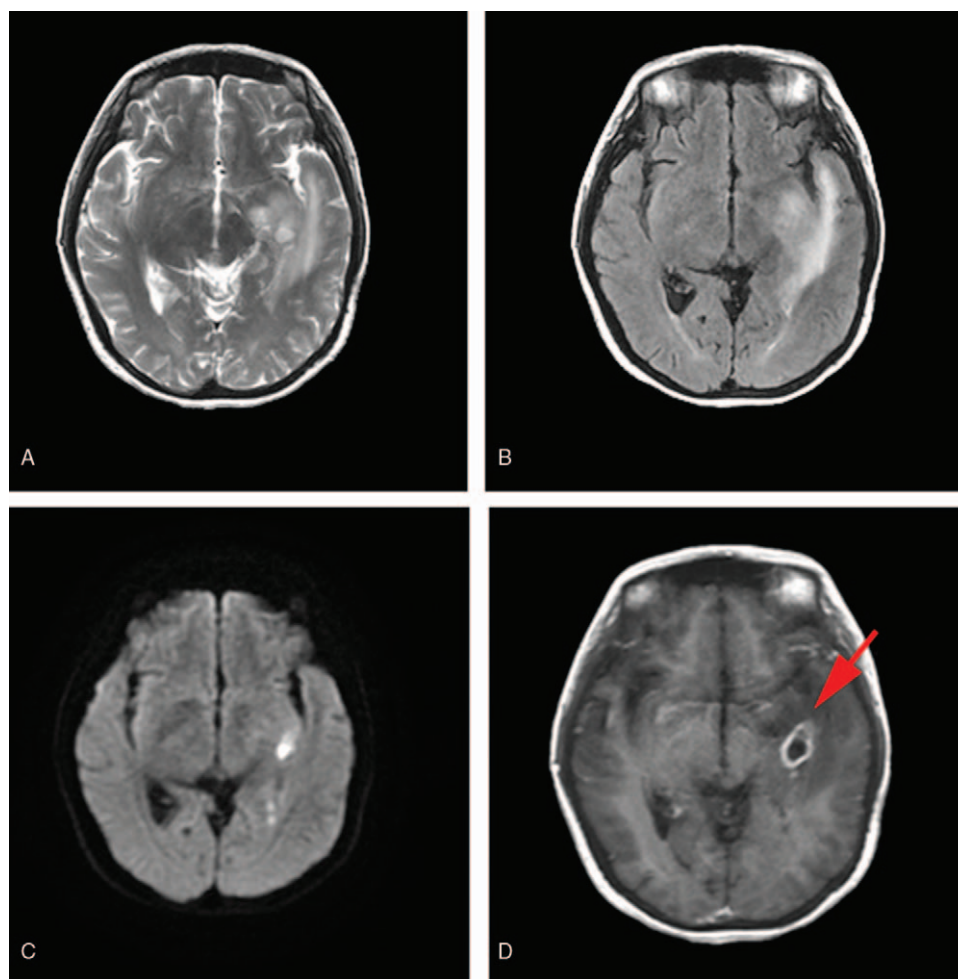


Figure 2. Head-enhanced MRI. (A) T2 axial view, (B) T2 FLAIR axial view, (C) diffusion-weighted imaging (DWI) axial view, and (D) T1 enhanced axial view of head demonstrating a ring-enhanced lesion (2.0×1.2 cm) (red arrow) in the left temporal brain with a large range of surrounding edema.

highly fastidious bacteria or fungi occurred. The first blood culture was performed at a local hospital, in which laboratory-based diagnostic techniques for defining fastidious or unusual pathogens may be unavailable. Secondly, penicillin G was used before referral to our hospital. During treatment, administration of antimicrobial agents reduced the recovery rate of bacteria by 35%–40%.^[3] Recurrent and repeated blood culture before antibiotic administration is critical for pathogenic evidence. Thirdly, true culture-negative endocarditis can be caused by uncommon or rare pathogens that show no growth in routinely used blood culture systems.

In this case, the infection source might be the painkiller injection spot on the skin for dysmenorrhea, suggesting *Dermatococcus* infection with effective antimicrobial therapy as counterevidence. However, intolerance to multi antibiotics increased the challenge regarding the choice of antimicrobial regimen. In terms of cross-reactivity, second- or third-generation cephalosporins may also be considered, as the degree of cross-reactivity with these agents and penicillin has been shown to be lower than that of first-generation agents. Three cephalosporins (cephazoline, cefuroxime, and ceftriaxone) with side chains differ from penicillin.^[5] Based on dissimilarity in chemical structures, the academy considered cross-reactivity between penicillin and second- or third-generation cephalosporins to be “highly unlikely.”^[6]

Antibiotics were changed several times until the cerebral abscess lesion almost faded. During this period, recurrent repeated echocardiogram showed the same vegetation without change. As the patient could not tolerate surgery, medical therapy would be appropriate for her. In this scenario, radiolabeled white blood cell single-photon emission computed tomography (SPECT/CT), which relies on the use of autologous radiolabeled leucocytes (^{111}In -oxine or $^{99\text{m}}\text{Tc}$ -hexamethylpropyleneamine oxime), is a specific method for detection of endocarditis and infectious foci.^[7] The radiolabeled white blood cell SPECT/CT could detect inflammatory activity in and around the vegetation as medical guidance for time to discontinue antimicrobial therapy. However, the patient refused due to economic dilemma.

The patient presented a single atrium with totally normal atrioventricular valves. Correction surgery should have been performed before developing to ES. However, she had missed the opportunity. Surgery indication for removal of vegetation was met (anterior mitral leaflet vegetation size ≥ 10 mm with mitral insufficiency^[3]). However, the process presented complexity not only due to cerebral abscess and ES but also because of the patient’s intolerance to cardiopulmonary bypass, with no benefits to experience for such a large project.

In summary, we reported a case of complex congenital heart disease (CHD) that developed into ES and complicated cerebral

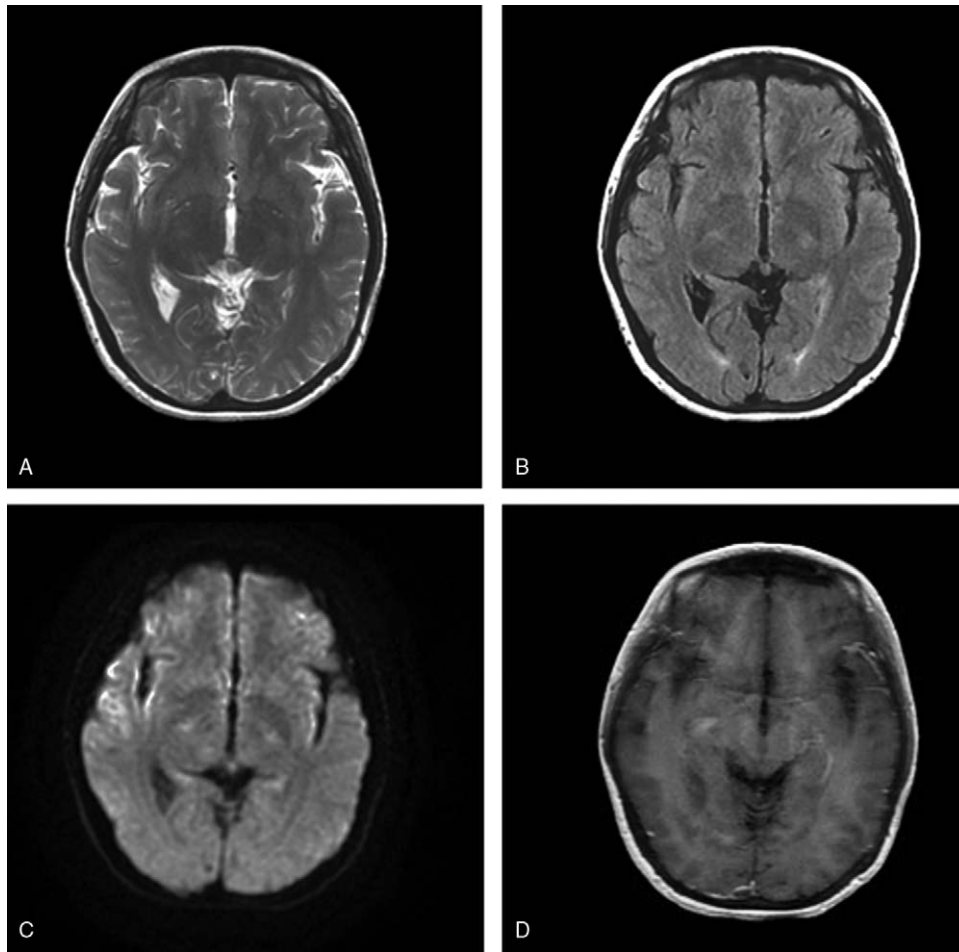


Figure 3. Repeated head-enhanced MRI after treatment. (A) T2 axial view, (B) T2 flair axial view, (C) DWI axial view, and (D) T1 enhanced axial view of the head demonstrating decreased ring-enhanced lesion in left temporal brain with a large range of surrounding edema.

abscess. Bidirectional intracardiac shunting (ES) occur when patients with complex CHD without correction surgery. In this scenario, infectious endocarditis, is prone to complicate in case of any infection from invasive interventions.

Author contributions

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