

Unexpected Infective Endocarditis in Corrected Congenital Heart Disease: A Case Report

Charlotte Johanna Cool, Rachmi Anasthasia, Aninka Saboe

Department of Cardiology and Vascular Medicine, Faculty of Medicine Universitas Padjadjaran
Dr. Hasan Sadikin General Hospital Bandung, Indonesia

Abstract

Objective: To describe patients with corrected Congenital Heart Disease (CHD) who experienced Infective Endocarditis (IE).

Methods: Two cases of IE were observed in 2019. The first case involved a 36-year-old woman with previous percutaneous transcatheter perimembranous ventricular septal defect (VSD) closure four months before admission. Echocardiography showed vegetation at noncoronary cusps of the aortic valve. Patient received antibiotics for six weeks and underwent surgery for evacuation of vegetation and device, along with VSD closure with a cardiovascular patch (Gore-tex). The second case involved a 43-year-old woman with a history of surgical closure in secundum atrial septal defect (ASD) by pericardial patch two months before admission. Echocardiography showed vegetation at the tricuspid valve. Patient received antibiotics for four weeks and planned for surgery to evacuate vegetation.

Results: IE is one of the major complications in CHD, whether uncorrected, treated, or corrected. The risk of IE increased with an invasive procedure. Post closure IE is rare. Poor dental hygiene and immunocompromised also increased patient's risk to be exposed to IE as shown in the first patient who had dental caries and the second patient who was on methylprednisolone for post-surgical pericardial effusion treatment.

Conclusion: The risk of IE increases with invasive procedures in CHD patients. Although the incidence of IE is quite rare, its possibility should become of a serious concern among physicians.

Keywords: Atrial septal defect, closure, infective endocarditis, ventricular septal defect

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Introduction

Congenital Heart Disease (CHD) is the most common underlying heart condition in Indonesia in patient with Infective Endocarditis (IE). Increased survival in children with CHD and the possible use of channels and prosthesis in corrective surgery contributes to an increase in the incidence of IE even though the patient has taken prophylactic antibiotics

in IE. However, CHD related IE mortality rates had decreased substantially to 10% due to improvements in IE diagnoses, antimicrobial treatment, cardiac surgery, and intervention therapy. Given the prognosis, morbidity, and expensive of IE management, IE prophylaxis has long been recommended in an effort to minimize the onset of IE.^{1,2}

CHD patient have a high risk of IE in their lifetime. One complication of IE remains a major concern in patient with CHD, whether uncorrected, treated, or corrected. The prevalence of endocarditis in an adult with CHD is 11 per 100,000 people per year higher than the general population that is 1.5-6 per 100,000 people per year. Mortality IE based on contemporary grown-up congenital heart disease (GUCH) still 16% and 1-year mortality

Correspondence:

Charlotte Johnna Cool,
Department of Cardiology and Vascular Medicine, Faculty of Medicine Universitas Padjadjaran-Dr. Hasan Sadikin General Hospital Bandung, Indonesia
e-mail: charlotte@unpad.ac.id

is 19%. Adults with repaired or palliated pulmonary atresia and corrected congenital transposition of the great arteries (TGA) have the highest incidence of IE with total cases 5.8 and 2.3 per 1000 patient-years. Patient with pulmonary stenosis (PS) or atrial septal defect (ASD) had a very low-risk incidence of IE, even in the adults with repaired or still patent arterial duct (PDA) IE non exists.^{2,5} Until now, very rarely IE reported in corrected CHD patients. In this case report, we discuss IE in patient with corrected CHD and what possible risk factors may underlie the occurrence of this condition.

Case Illustration First Case

First case, a 36 year-old Asian woman, a teacher, came to our emergency department with chief complaint weak left limb three days before hospitalization with lips deviation and slurred speech. She didn't complaint severe headache and projectile vomit. There was a history of dyspnea on the effort since childhood.

There was no history of fever before. She complained of painful in legs two months before being hospitalized and resolved with pain medication. The patient complained that there are reddish spots on the palms and soles of the feet 1 month before admission, she also complained of a lump in the tips of the fingers that felt painful one month before being hospitalized. The patient has never experienced reddish lines on the nails. The patient never experiences redness in the

eyes. Her complaints of reddening urination without pain which is accompanied by pain when urination is denied. Patient did not complain about a decrease in consciousness. There were cavities that have only been known in the past two months before being hospitalized, but the patient did not go to the doctor. The patient was known as a patient with congenital heart disease since childhood. Previously, she underwent corrected VSD with Amplatzer duct occluder (ADO) 4 months before hospitalization.

In our emergency room, the patient looks moderately ill, fully alert. The blood pressure was 80/50 mmHg, 75 x/minutes of resting heart rate, 20 x/minutes of respiratory rates, and normal temperature 36,7°C. In physical examination showed jugular vein pressure normal. The thoracic examination showed there is no cardiomegaly, and there is no additional heart sound. There was pansystolic murmur with grade 3/6 in linea left sternalis border with Carvallo sign negative, and pansystolic murmur with grade 3/6 in apex referred to the axilla. Other physical examinations were unremarkable. In neurology examination showed positive light reflex in both eyes; there was no Roth spot's, eyeball movements were normal. There was a history of the parenthesis of central nerve VII and left nerve, XII. In motoric examination were found 5 in the right upper and right lower extremity and in the left upper and left lower extremity the motoric were 0. The physiologic reflex score was +2/+2 and pathologic reflex negative.

The laboratory examination showed

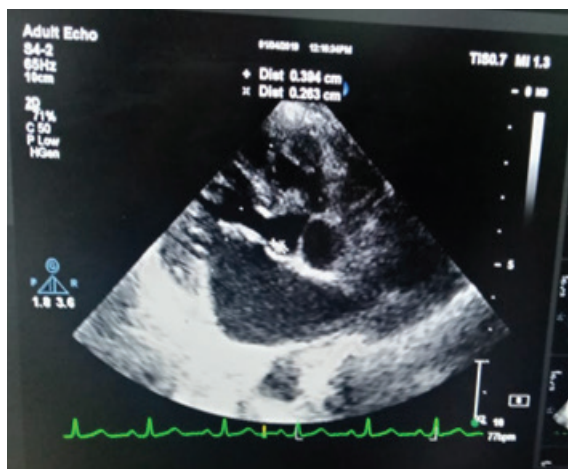


Fig. 1 Echocardiography Showed Vegetation at NCC of the Aorta

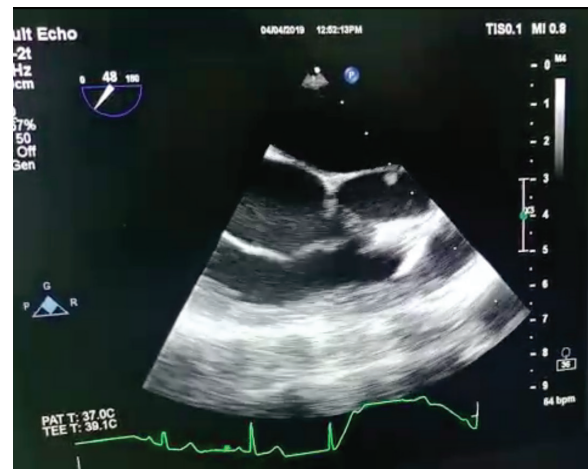


Fig. 2 Transesophageal Echocardiography Showed Multiple Vegetation Attach of the Tip of Aortic Valves

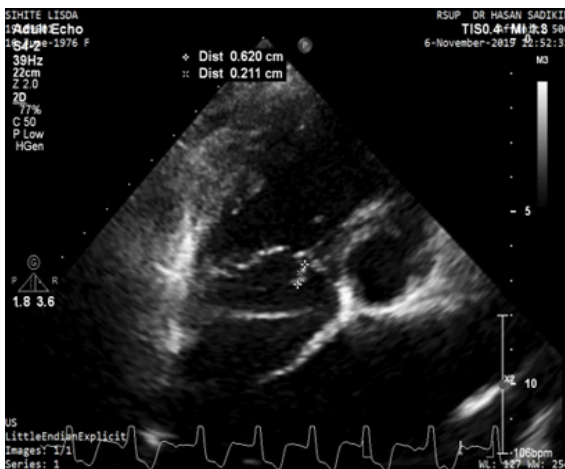


Fig. 3 Echocardiography Showed Vegetation at Tricuspid Valve



Fig. 4 Echocardiography Showed Vegetation Size was Increased

anaemia and other examination within normal limits. Electrocardiography, chest X-Ray, and Other examination within normal limits.

The patient was diagnosed on the emergency department with stroke infarct dd stroke haemorrhage, congenital heart disease VSD post transcatheter closure, *Possible Infective endocarditis. (Duke Criteria: 3 Minor criteria: Predisposing heart Condition (VSD) Osler node, Janeway lesion)*. The patient was given antibiotic empiric with adjusted dose ceftriaxone 1x2 g iv dan gentamicin 3mg/kg/48 hour iv, aspilet 1x81 mg PO, paracetamol 3x 500 mg PO.

Blood culture was taken three times with a time interval between first examinations and last examination 1 hour. Results of culture showed *Enterococcus sp* which sensitive to all antibiotics except tetracycline.

Patient undergoing Echocardiography (Fig.1) examination on March 1, 2019, showed VSD perimembranous (PM) s/p device closure with suspect additional VSD PM Left to Right shunt, Moderate PS due to protruding device, Vegetation at NCC of Aorta.

Patient undergoing transesophageal echocardiography (TEE) (Fig.2) examination on March 4, 2019, showed VSD PM s/p closure, residual shunt (-); Moderate PS due to protruding part of the device (right ventricle (RV) part); small, mobile, multiple vegetation attach of the tip of aortic valves; Mild AR, eccentric jet due to mal-cooptation of aortic valves.

The patient then diagnosed with Definite Infective Endocarditis, VSD perimembranous with complication suspected of cardioembolic

stroke infarction (Duke Criteria: 1 major criterion: Echocardiography positive for IE (vegetation), 3 Minor criteria: Predisposing heart Condition (VSD) Osler node, Janeway lesion), suspect additional VSD perimembranous.

The patient was given antibiotic treatment Vancomycin 3x500 mg iv for four weeks. During the treatment, there were no complications. After two weeks of therapy, the patient was taken blood culture two times with negative culture results, and antibiotic therapy continued until six weeks. After 17 days of treatment, the patient has re-evaluated echocardiography and found that vegetation disappeared. The patient then scheduled for surgery for evacuation of vegetation; the device removes and VSD closure. The patient underwent surgery on the 30th day of treatment, and an intra-operative finding was found: dislocated aplazter partially, Supracristal or Doubly Committed (SADC) position VSD, closure of VSD with Gore-tex, vegetation (-).

Second Case

Second case, a 43-year old Batak woman, a house wife, came to our emergency department with palpitations. There was no history of fever before. She never experienced pain in legs, reddish spots on the palms and soles of the feet, a lump in the tips of the fingers that feels painful, reddish lines on the nails, redness in the eyes, reddening of urination without pain or decrease in consciousness. The patient was known as a patient with congenital



Fig. 5 Echocardiography Showed Multiple Vegetation with Various Size

heart disease. Previously, she already had corrected ASD with a pericardial patch (Gore-tex) 2 months before hospitalization. She also had a pericardial window because of severe pericardial effusion one months before admission, and she took corticosteroid 24 mg per day since one months before admission.

In physical examination, the patient looks moderately ill, fully alert. The blood pressure was 110/70 mmHg, 68 x/minutes irregularly irregular of resting heart rate, pulse was 60 beats per minutes irregularly, unequal, 20 x/minutes of respiratory rates, and normal temperature 36,8°C. In physical examination showed jugular vein pressure normal. The thoracic examination showed there are cardiomegaly and variable of heart sounds. There was pansystolic murmur with grade 3/6 in linea left sternalis border with Carvallo sign positive. Other physical examinations were unremarkable. In neurology examination showed positive light reflex in both eyes; there was no Roth spot's, eyeball movements were normal.

The laboratory examination showed anaemia and other examination within normal

limits. Chest X-Ray showed cardiomegaly with the sign of pulmonary hypertension. On electrocardiography showed atrial fibrillation with right axis deviation and right ventricle hypertrophy.

Patient were taken blood culture three times with a time interval between first examinations and last examination 1 hour. Results of culture showed *Acinetobacter sp* which sensitive to all antibiotics except cotrimoxazole.

She underwent TEE (Fig.3) one day after admission, with the results ASD s/p surgical closure, multiple residual shunts, vegetation at tricuspid valve with size (2x6 mm).

The patient was diagnosed with ASD post-surgical closure with a residual shunt, AF NVR, Possible Infective endocarditis. (Duke Criteria: 1 Major criterion: echocardiogram showed vegetation, 1 Minor criterion: Predisposing heart Condition (ASD)). The patient was given antibiotic empiric with adjusted dose Ampicillin Sulbactam 4x3 gr iv and Gentamycin 1x160 mg iv.

The patient was given antibiotic treatment Ampicillin Sulbactam 4x3 gr iv and Gentamycin

1x160 mg iv for four weeks. During the treatment, there were no complications. After two weeks of therapy, the patient was taken blood culture two times with negative culture results, and antibiotic therapy continued until six weeks. After 22 days of treatment, the patient has re-evaluated echocardiography (Fig.4) and found vegetation increases in size became 3x3 mm. After 34 days of treatment, the patient has re-evaluated echocardiography (Fig.5) and found multiple vegetation with various size from 3.9x9.4 mm until 7x3 mm.

In 28 days of treatment, she got urticaria in her body, and she was found suspect allergic from ampicillin and gentamycin, so the antibiotic was replaced with ceftriaxone 2x2 gr iv in 28 days of treatment. The patient then scheduled for surgery for evacuation of vegetation, double valve replacement and ASD repair.

Discussion

Bacteremia following cardiac catheterization is rare. In a prospective study from Banai, etc., they evaluated 960 patient undergoing catheterization, only 0.4% of patient had positive blood cultures that significantly associated with cardiac catheterization. In the literature, there were 8 cases of IE after catheterization. From the 8 cases, there were found four patient with a history of valve disease and IE occurred after catheterization. Signs and symptoms of catheterization are obtained from day one until one week after the procedure.^{3,6-9}

Risk factor for IE is patient with prosthetic cardiac valves, IV drug users, patient with mitral valve prolapse (MVP) or non-rheumatic heart disease (CHD, bicuspid aortic valves), rheumatic heart disease. The risk of IE increases in patient who undergo invasive procedures. Patient with old age, alcoholics, sufferers of chronic inflammatory bowel disease, poor dental hygiene, repeated dialysis, diabetes mellitus, and patient with immunosuppressants are at risk to be exposed from IE.^{5,10}

Patient with a moderate risk of developing IE include patient with PDA, bicuspid aortic valve, and coarctation of the aorta, rheumatic heart disease (RHD) or hypertrophic obstructive cardiomyopathy (HCOM). Adults with CHD (GUCH), including groups with low-risk factors.^{3,11}

Patient with a high risk of developing IE include patient who have prosthetic heart valves, a history of previous endocarditis,

cyanotic CHD, surgically constructed systemic pulmonary shunts or conduit. Patient with prosthetic valves have 5-10 times the risk compared with native valves.^{3,11}

IE related to post-closure is a rare case, but there are several case reports. There is one case report from Minnesota with one 66-year-old male with a large ASD that has been catheterized with a 32-mm ASD Amplatzer occlude 30 months before the incident. A case report from India also discusses the same thing. In the first case, the treatment was done by surgery, and the patient received IV antibiotic Vancomycin for six weeks while the second patient was given Daptomycin IV for six weeks. From these two patient, significant clinical signs and outpatient were obtained after six weeks. There is one case report of IE on VSD after closure reported by Schuerman in 2005 in a 22-month-old baby girl. She is experiencing a fever one month after the closure that lasted for one month and did not respond by giving antibiotics.¹²⁻¹⁵

In the first case, we found several risk factors for IE namely VSD that has high risk developing of IE, and patient had undergone the invasive procedure and this increase the risk of IE. She also had cavities which is one of a risk factor for IE. Maintaining good oral hygiene and infection control can decrease the incidence of IE. It should be emphasised to patients that maintaining optimal oral health and hygiene and regular dentist visits may reduce the incidence of bacteraemia from brushing teeth, chewing food and daily activities, and thus are particularly important in reducing the risk of IE. In the second case, we consider the risk factor of IE that patient had immunocompromised state because she took corticosteroid for a long time beside she already had CHD.^{15,16}

Patient with a high risk of IE should be given prophylaxis before medical treatment. Prophylactic antibiotics only needed if the patient undergoes the invasive procedure. In action in the field of cardio, prophylactic antibiotics can be given to patient who will undergo prosthetic valve implantation or pacemaker installation. Prophylactic antibiotics must be considered in patient who will undergo dental procedures. At-risk procedures involve manipulation of the gingival or periapical region of the teeth or perforation of the oral mucosa (including scaling and root canal procedures). Antibiotic prophylaxis should only be considered for patient at highest risk for endocarditis.¹⁷

Prophylactic antibiotics must be started

as soon as possible before the action starts and stops after 48 hours post-procedure. The recommended prophylactic antibiotic dose in patient with high risk are amoxicillin or ampicillin 2 g oral or intravenously. If the patient has an allergy to penicillin or ampicillin, 600 mg of oral or intravenous clindamycin can be given.¹⁶

Management of IE should be started promptly. In ESC 2015 about IE we can give empirical treatment in patient whose microorganisms have not yet been identified. Once the pathogen is identified, the antibiotic treatment must be adapted to its antimicrobial susceptibility pattern.¹⁷

In the first case, the patient has been given ceftriaxone 1 x 2 g iv and gentamicin 3 mg/kg / 48 hours iv, and in the second case, the patient has given Ampicillin Sulbactam 4x3 gr iv and Gentamycin 1x160 mg iv. As we know in 2015 European Society of Cardiology (ESC) guidelines about IE, in patient with unknown types of bacteria can be given gentamicin 3 mg/kg/hour iv. In the guideline mentioned, gentamicin administration can be given with vancomycin, but the patient did not get vancomycin but get ceftriaxone at the emergency room due to unavailability of drugs. Patient get vancomycin 3x500 mg iv when entering the ward.

After the results of the culture came out, we found the bacteria that causes IE in these patient was *Enterococcus spp.* Based on the 2015 ESC guidelines on IE, there are three choices of IE antibiotic therapy regimens caused by *Enterococcus spp.* i.e. Amoxicillin 4 x 200 mg / kg / day iv plus gentamicin 1x 3 mg / kg / day intravenous (IV) or intramuscular (IM) for 4-6 weeks or can be given ampicillin 4 x 200 mg / kg / day iv plus ceftriaxone 2x4 gr / day iv for 6 weeks or Vancomycin 2x 30 mg / kg / day plus Gentamicin 1 x 3 mg / kg / day given for 6 weeks.

Valve surgery is fundamental in the standard of care of selected IE patient. Surgical intervention can be a lifesaving procedure in

complicated, critically ill endocarditis patient. Heart failure, uncontrolled and persistent infection despite optimal antibiotic therapy and recurrent emboli are the major indications for valve surgery in IE population.¹⁷⁻¹⁸

In the first case, the patient had a complication that is acute stroke. The timing valve surgery in IE patient with stroke remains controversial. Stroke is an independent risk factor for postoperative mortality in IE patient. One clinical quandary is whether early valve surgery can be safely performed within seven days after a stroke or if it is better to postpone surgery for at least one week. No randomized trials have addressed this conundrum. Investigations have suggested a better outcome for IE patient with ischemic stroke who undergo early cardiac surgery. Ruttman *et al.* analyzed 65 patient who underwent cardiac surgery after cardioembolic (embolic) stroke (median time, four days; range, 0-38 days). Surgery in this time frame was not associated with worse patient outcomes.¹⁸

In the second case, the patient had locally uncontrolled infection marked by enlarged vegetation size and multiple vegetation in 22 days of treatment. Uncontrolled infection is most frequently related to a perivalvular extension or "difficult-to-treat" organism. Unless severe co-morbidity exists, the presence of locally uncontrolled infection is an indication for early surgery in patient with IE.¹⁷

In this case report, there were two women with CHD post corrected and experienced IE with a different risk factor. From the first case, the risk factor was cavities and didn't go to the dentist; meanwhile, in the second case, the risk factor was immunocompromised because she took methylprednisolone for treatment pericardial effusion. Although the incidence is rare, the possibility of IE must be a concern for a physician, especially for those who have other clinical condition like poor dental hygiene and immunocompromised.

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