Case Report

Infective endocarditis of transcatheter atrial septal occluder devices: A case report

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Abstract

Infective endocarditis (IE) is caused by bacterial vegetation in valves, but it can also occur in implanted mechanical devices. We report a rare case of IE occurring at the site of percutaneous atrial septal closure devices in a patient in her 50s that had been placed for residual defects on a closure patch in her childhood for an atrial septal defect (ASD). She also had a medical history of distal pancreatectomy for insulinoma in her 40s and had insulin-dependent diabetes mellitus, which means she had been immunocompromised.

She visited our hospital with complaints of fever and lumbar pain. A computed tomography scan revealed liver abscess. In blood, urine, and drainage specimens submitted for culture testing, extended spectrum beta-lactamase-producing Escherichia coli was cultured in all specimens. Echocardiography showed vegetation at the atrial septal closure devices. In accordance with IE therapy, removal of the atrial septal patch and closure device was performed after antibiotic treatment for 6 weeks.

Because the atrial septal patch was calcified and the two devices implanted on the patch were not well covered by neointima, bacteria could easily form vegetation. Percutaneous residual ASD closure on an atrial patch, especially for immunocompromised hosts, should be carefully considered.

Learning objective: In general, neointima forms and coats a closure device several years after its insertion. However, as in the present case, the closed atrial septal patch may be severely calcified and the neointima may not be sufficient to form on the closure device, and infective endocarditis may occur at the site of implantation. In some cases, the indication for closure device implantation after atrial septal patch closure should be carefully considered.

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Introduction

In recent years, the number of cases of cardiac device-related infective endocarditis (CDRIE) has been increasing with increase in the use of pacemakers and other devices. The increase in the number of cases of CDRIE may be attributed to the recent increase in procedures using artificial devices (pacemaker leads, catheters, stents, etc.) as well as artificial valves and also the aging of the population [1]. The proportion of CDRIE cases, excluding local infections, is approximately 10% to 23% of all device-related infections [2,3].

It is recommended that antimicrobial agents be selected according to the susceptibility of the isolated organism and be administered for approximately 6 weeks in cases of CDRIE. As with treatment for refractory Gram-negative bacterial infections, a combination of amikacin and gentamicin has been used; however, an appropriate treatment regimen, including the duration of treatment with the combination, has not been determined. In many cases, treatment with antimicrobial agents alone is difficult and early surgery should be considered. However, surgical device removal with open heart surgery is associated with a high risk of perioperative mortality [4].

We experienced a case of IE that developed at the site of insertion of an endocardial device in a patient who had undergone patch closure for an atrial septal defect (ASD) in her childhood and transcatheter atrial septal closure for residual defects several years previously. Although the antibiotic susceptibility of the causative organism in this case,
extended spectrum beta-lactamase (ESBL)-producing *Escherichia coli*, was good, IE caused by Gram-negative rods was considered to have a poor prognosis. CDRIE may recur if an infected intracardiac medical device is left behind after insertion. Complete removal of the device is recommended in the 2015 European Society of Cardiology guidelines for CDRIE [5] and aggressive source control treatment was performed in this case in accordance with the guidelines.

**Case report**

A female in her 50s had a history of patch closure for ASD in her childhood. Several years previously, two residual shunts after patch closure for ASD were treated with placement of a Cribriform Occluder 25 mm (St. Jude Medical [now Abbott], St. Paul, MN, USA) [residual shunt on the side of the aorta and superior vena cava (SVC)] and an Amplatzer Septal Occluder 15 mm (St. Jude Medical [now Abbott], St. Paul, MN, USA) [residual shunt on the side of the inferior vena cava (IVC)]. In addition, she had a medical history of distal pancreatectomy for insulinoma in her 40s. Laboratory data showed insulin-dependent diabetes mellitus, but insulin therapy was not initiated until her admission.

The patient came to our hospital with a chief complaint of fever and lumbar pain and was admitted to the Department of Gastroenterology on the same day because an abdominal computer tomography scan, which was performed on day 1, showed acute nephritis and liver abscess (Fig. 1).

Four sets of blood and urine cultures were submitted on the day of admission. Percutaneous trans-hepatic abscess drainage was performed the day after admission, and antibiotic therapy was started. At first, she was treated with antibiotics. Ampicillin-sulbactam was administered by intravenous infusion on day 2 to day 3. The antibiotic combination was changed to cefmetazole from day 4 to day 6 due to the detection of Gram-negative rods in the culture test. The causative organism was confirmed to be ESBL-producing *E. coli*, which was detected from all samples, and the antibiotics were therefore changed to meropenem and gentamicin from day 7.

Transthoracic echocardiography was performed as screening on day 5 and it showed no vegetation on the native valve, but there was a mobile vegetation measuring 18.0 × 6.3 mm on the left atrial side of the Cribriform Occluder. In addition, 6.5 × 10 mm vegetation with a 10-mm-long stalk-like structure was found on the right atrial side of the Amplatzer Septal Occluder (Online Fig. 1). She was transferred to the Department of Cardiology on day 5 due to complications of endocarditis. Transesophageal echocardiography (TEE) performed on day 9 showed an 8-mm-long cord-like vegetation on the left atrial side of the Cribriform Occluder and a 10-mm-long cord-like vegetation on the right atrial side of the Amplatzer Septal Occluder (Fig. 2). There was neither abnormal blood flow adjacent to the two devices nor residual shunt flow on the atrial septum, but there was a low echoic area between her atrial septum patch and Cribriform Occluder. There was no vegetation on her native valves. On the same day, brain magnetic resonance imaging showed no evidence of cerebral infarction.

Blood cultures were performed on day 10 and were negative. On day 25, TEE was performed again, and the vegetations had become too small to measure. Laboratory data were almost in normal ranges, and she had no fever after day 7. However, the low echoic area, which was considered to be an abscess or edema, was still present.
Because the vegetations were becoming smaller and no embolism, including pulmonary thrombosis, was detected, IE was determined not to be treatment-resistant. Therefore, we opted for a wait-and-see procedure. Atrial septal reconstruction (patch removal, Cribriform Occluder and Amplatzer Septal Occluder removal) was performed on day 53 after about 6 weeks of treatment with antibiotics. The center of the Amplatzer Septal Occluder on the SVC side showed vegetation, and the occluder was removed after removal of the vegetation and detachment of the closure patch. The closure patch was confirmed to be a dacron fabric sheet. The septal closure patch was incised to the site of the Cribriform Occluder on the IVC side. After dissecting the neointima of the right and left atria, the Cribriform Occluder was removed and all remaining dacron fabric sheets were removed. The defect was sutured closed with the autologous pericardium that had been harvested because the defect hole was 50 × 20 mm (Fig. 3A). Examination of the surgically removed specimen showed marked calcification of the dacron fabric sheet. Although neointima had formed between the atrial septal closure patch and the Amplatzer Septal Occluder and Cribriform Occluder, both devices were not well covered (Fig. 3B). After the surgery, the patient was discharged on day 77 without any complications.

Discussion

There are several reports of IE occurring after placement of an atrial septal defect patch alone and Amplatzer Septal Occluder alone [6,7]. However, to the best of the authors’ knowledge, this case is a rare case of IE occurring after implantation of an Amplatzer Septal Occluder and a Cribriform Occluder for residual shunts after patch closure. Staphylococcus aureus is the most common causative organism of CDRIE, accounting for more than half of all CDRIE cases. Coagulase-negative staphylococci are particularly common [8]. Device infections may be mixed infections caused by multiple causative organisms, and care should be taken to identify the causative organism [9].

ESBL-producing E. coli are rarely identified as the causative organism of CDRIE from various culture tests in the clinical course of a long period such as a period of several after decades of use of a patch closure or a period of several years of use of an atrial septal closure device. Risk factors for CDRIE include renal failure, use of a steroid, congestive heart failure, hematoma formation in the pocket of a device, diabetes mellitus, and use of an anticoagulant [10]. Our patient had suffered from an insulinoma-producing endocrine tumor of the pancreas 15 years previously and had undergone distal pancreatectomy, resulting in insulin-dependent diabetes mellitus. It is assumed that this made her an immunocompromised host.

Judging from the various cultures and the clinical course of our case, it is thought that ESBL-producing E. coli had invaded the blood vessels through a urinary tract infection. Because she was immunocompromised, her infection might have led to bacteremia. In addition, her distal pancreas had been resected, which means the bacteria could easily enter the blood stream and could form a liver abscess and vegetation on the devices.

The atrial septal patch, which was implanted in her childhood, was markedly calcified and probably not adequately covered by neointima, resulting in uncontrolled residual shunts. The residual shunts were managed after implantation of an Amplatzer Septal Occluder and a Cribriform Occluder several years previously. However, the neointima between the atrial septal closure patch with prominent calcification and the atrial septal closure device did not form well even in the chronic phase after implantation, possibly contributing to the susceptibility to IE.

The 2015 European Society of Cardiology guidelines [5] recommend total removal of medical devices, and all medical devices were removed during the re-cardiac surgery. In this case, we decided to perform
(A) The two Amplatzer devices (Cribriform Occluder and Amplatzer Septal Occluder) were identified from the right atrial view of the incision. Vegetation was found in the center of the Amplatzer Septal Occluder on the superior vena cava side, and after removal of the vegetation, the neointima that had formed on the right atrial side was detached. The patch closure corresponding to the original fossa ovalis was incised caudally. The patch used in the previous procedure was found to be a dacron sheet because the material was fabric. After securing the left atrial view, the left atrial mesh part was also removed and the Amplatzer Septal Occluder was removed. Furthermore, the septal closure patch was incised to the site of Cribriform Occluder implantation. The neointima that had formed between the patch and the Cribriform Occluder was peeled off and the device was removed. The surrounding tissue was dissected and all remaining dacron sheets were removed. The defect hole was approximately 50 × 20 mm. The atrial septal defect was sutured closed with autologous pericardium while reinforcing the margins with bovine pericardium from the inferior vena cava side.

(B) The patch that was implanted for an atrial septal defect in her childhood showed marked calcification. There was neointima between the closure patch and the atrial septal closure device; however, there was not sufficient neointima formation to adequately cover the closure device.

(a) Image of the left atrial side of the Cribriform Occluder implanted in the inferior vena cava.
(b) Image of the right atrial side of the Amplatzer Septal Occluder implanted in the aortic site.
closure with autologous pericardium in order to prevent recurrence of CDRIE and new onset of heart failure.

In the present case, atrial septal closure devices implanted on an atrial septal patch with significant calcification were not well covered with neointima. In addition, we experienced a rare case of bacteremia that developed from a urinary tract infection caused by ESBL-producing E. coli in an immunocompromised host and in whom CDRIE developed in the chronic phase after implantation and was cured by surgical treatment.

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CRediT authorship contribution statement

The authors contributed to the present study as follows.
Mirei Nabuchi-Kasawaki, MD, a principal investigator of this study, participated in the study design, collection of patient data, and analysis of data and drafted the initial version of this manuscript.

Takahiro Doi, MD, PhD, Tomohiro Mita, MD, PhD, Syunsuke Sasaki, MD, PhD, Mitsugu Hirokami, MD, PhD, and Shuichi Naraoka, MD, PhD, participated in the study design and the collection and analysis of clinical data.

Satoshi Yuda, MD, PhD, FJCC, contributed to the study conception and design and assisted with integration of the data into the manuscript.

Declaration of competing interest

None declared.

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