

# Staged transcatheter closure for atrial septal defect and patent ductus arteriosus: a case report

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## Background

Atrial septal defect (ASD) and patent ductus arteriosus (PDA) are both common congenital heart diseases, but the combination of these two cardiac defects is extremely rare, and the therapeutic strategy is controversial.

## Case summary

We treated an adult patient with combined ASD and PDA, and safely attained a successful outcome with two-stage transcatheter closure, which is PDA closure preceding ASD closure, to prevent serious complications.

## Discussion

Transcatheter closure of one of the defects is now widely accepted as an alternative to surgical closure. In addition, adults with both ASD and PDA are better suited for transcatheter closure than surgical closure. One of the reasons is the difficulty to ligate the ductus arteriosus of an adult patient due to its friability and calcification. Meanwhile, simultaneous combined transcatheter closure of ASD and PDA can result in serious complications, such as thrombocytopenia and haemolysis, whose mechanism is considered to be the destruction of platelets and red blood cells by the residual shunt through implanted devices. Additionally, antiplatelet therapy that prevents device-related thrombus formation after ASD closure can possibly exacerbate thrombocytopenia and haemolysis. Therefore, the staged strategy is reasonable from the perspectives of antiplatelet therapy and haemodynamic changes.

## Keywords

Congenital heart disease • Atrial septal defect • Patent ductus arteriosus • Case report

## Learning points

- The combination of atrial septal defect (ASD) and patent ductus arteriosus (PDA) is extremely rare, and it is reasonable that transcatheter closure for PDA should precede ASD closure from the perspectives of antiplatelet therapy and haemodynamic changes.

## Introduction

Atrial septal defect (ASD) and patent ductus arteriosus (PDA) are both common congenital heart diseases, but the combination of these two common cardiac defects is extremely rare.<sup>1–3</sup> Transcatheter closure of these defects is widely accepted as an alternative to surgical closure. Previously, we reported a rare case of an adult patient with both ASD and PDA and subsequently underwent simultaneous combined transcatheter closure.<sup>4</sup> The case was

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followed by severe thrombocytopenia and subclinical haemolysis, thereby prompting us to discontinue antiplatelet agents that had been prescribed to prevent device-related thrombus formation after ASD closure. Therefore, we need to reconsider the transcatheter strategy for a patient with the combination of ASD and PDA.

## Timeline

Time	Events
2016~	The patient had dyspnoea upon exertion [New York Heart Association (NYHA) Classification II]
June 2017	Transthoracic echocardiography (TTE) showed a secundum-type atrial septal defect (ASD) at her previous hospital
July 2017	She was referred to our hospital and TTE showed not only ASD but also patent ductus arteriosus (PDA)
September 2017	Transcatheter closure of the PDA using Amplatzer Duct Occluder, the Qp/Qs ratio decreased from 4.15 at baseline to 2.11
January 2018	Transcatheter closure of the ASD using Amplatzer Septal Occluder/started antiplatelet therapy
February 2018~	She exhibited no dyspnoea upon exertion (NYHA Classification I)

## Case presentation

A 36-year-old woman with dyspnoea upon exertion [New York Heart Association (NYHA) Classification II] presented at the referring hospital. Transthoracic echocardiography (TTE) showed a secundum-type ASD with dilatation of both the right atrium (RA) and the right ventricle (RV), as indicated by a Qp/Qs ratio of 2.40. She was referred to our hospital for transcatheter closure of the ASD. A systolic murmur was heard at the left sternal border of 2nd intercostal space, which implied relative pulmonary valve stenosis. Cyanosis and clubbed finger were not observed. Her chest radiograph showed cardiomegaly and protrusion of the left second bow. Transthoracic echocardiography performed at our hospital showed not only ASD but also irregular shunt flow in the pulmonary artery (PA), thereby indicating PDA. The transoesophageal echocardiography showed a secundum ASD with a diameter of 19 × 14 mm, and there was a sufficient rim around the defect (Figure 1A and B). In addition, reconstructed three-dimensional computed tomography revealed a PDA [Krichenko Type A (Figure 1C and D)]. The diameters of the PA side and aorta (ampulla) side were 2.5 and 9.7 mm, respectively. The duct length was 6.9 mm. Because simultaneous closure of both the PDA and ASD could lead to serious complications, such as thrombocytopenia or haemolysis, augmented by antiplatelet therapies to prevent device-related thrombus formation after ASD

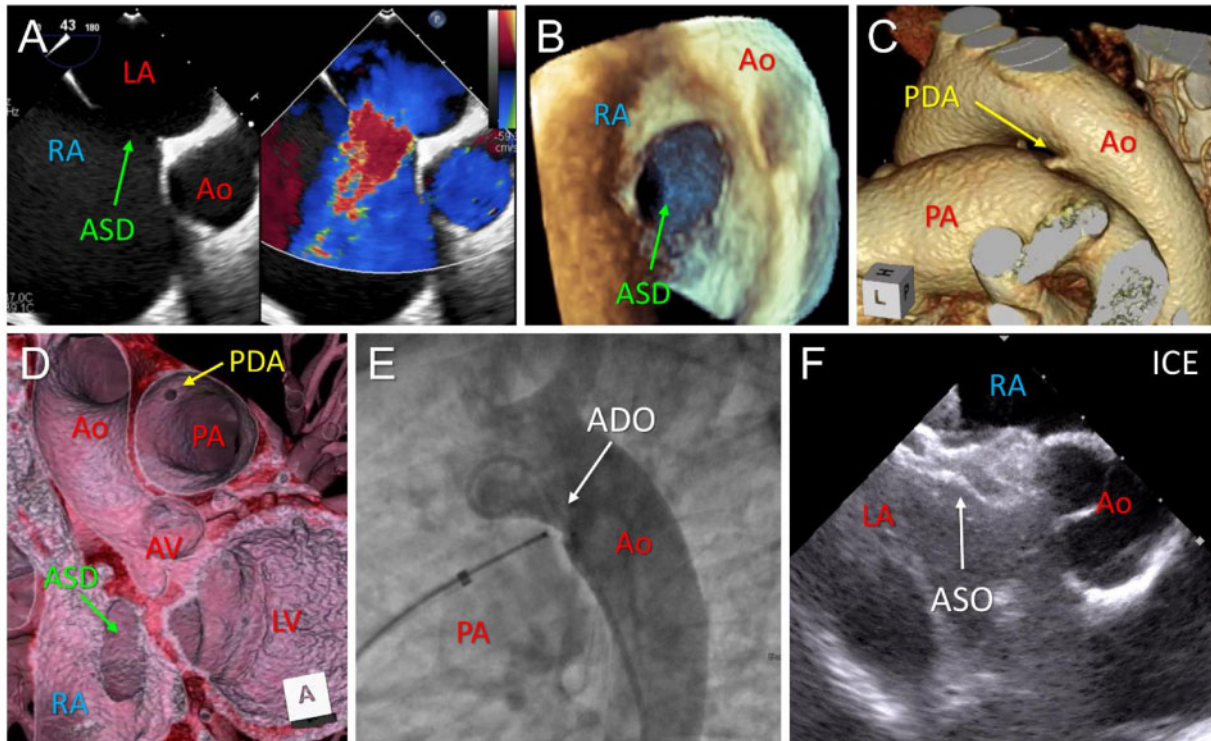
closure, PDA closure was performed before ASD closure. After the transcatheter closure of the PDA using a 6/4 mm Amplatzer Duct Occluder (ADO) (Abbott Vascular, Abbott Park, IL, USA), the Qp/Qs ratio decreased from 4.15 at baseline to 2.11, as measured by cardiac catheterization (Figure 1E). The mean PA pressure decreased from 22 mmHg to 16 mmHg on right heart catheterization before and after PDA closure. Echocardiography revealed an estimated PA systolic pressure of 31 mmHg before PDA closure, which decreased to 23 mmHg 3 months after the procedure. Four months after PDA closure, no residual shunt was observed in a subsequent TTE, and then, we successfully completed a transcatheter closure of ASD with a 19 mm Amplatzer Septal Occluder (ASO) (Abbott Vascular, Abbott Park, IL, USA) using intracardiac echocardiography imaging guidance (Figure 1F). There was no residual shunt through the ASD, with a decrease in size of both the RA and RV. She exhibited no dyspnoea upon exertion (NYHA Classification I) after the completion of both the PDA and ASD closures.

## Discussion

Atrial septal defect and PDA are both common congenital heart diseases; however, the combination of the two defects is considered extremely rare, with a frequency of 0.08% to 1.20% among patients with congenital heart diseases.<sup>1–3</sup> The combination of these two common cardiac defects might be rare because of reduced PDA flow in the foetus resulting from shunting from the RA to the left atrium (LA) through a large ASD, which promotes PDA closure. In addition, the oxygen concentration in the PA had increased following shunting from the LA to the RA through the ASD after birth, which also promotes spontaneous closure of the PDA.<sup>2</sup> In the foetus, the blood in the ductus arteriosus flows from the PA to the aorta. After birth, the neonate starts to breathe using the lungs, and the pulmonary vascular resistance decreases. Consequently, the blood flow through the ductus arteriosus becomes bidirectional, and then flows only from the aorta to the PA.<sup>5</sup> If the patient has ASD, the oxygen concentration in the ductus arteriosus is high earlier after birth because the blood oxygenated by the lungs flows from the LA to the RA through the ASD and goes from the PA to the aorta through the ductus arteriosus.

Transcatheter closure of one of the defects is now widely accepted as an alternative to surgical closure. A paediatric patient who had both ASD and PDA was successfully treated by combined transcatheter closure using an ASO for ASD and an ADO for PDA.<sup>6</sup> Although surgical closure is also the standard option for ASD and/or PDA, it is sometimes difficult to ligate the ductus arteriosus of an adult patient due to its friability and calcification. Adult patients with PDA might be better suited for transcatheter closure with either occlusion devices or coils because of its high success rates and few complications.<sup>7</sup>

Previously, we reported a rare case of an adult patient with both ASD and PDA and subsequently underwent simultaneous combined transcatheter closure.<sup>4</sup> The case was followed by severe thrombocytopenia and subclinical haemolysis, suggested that total bilirubin and lactate dehydrogenase were elevated albeit the haemoglobin level remained within normal limits and urinalysis showed no haemoglobinuria. Transthoracic echocardiography confirmed a residual



**Figure 1** Evaluation of atrial septal defect and patent ductus arteriosus by echocardiography, reconstructed three-dimensional computed tomography, and angiography: (A and B) atrial septal defect observed on transoesophageal echocardiography (green arrow); (C) assessment of patent ductus arteriosus by the three-dimensional computed tomography (yellow arrow); (D) combined atrial septal defect (green arrow) and patent ductus arteriosus (yellow arrow) observed on reconstructed three-dimensional computed tomography. (E) Amplatzer Duct Occluder (white arrow) deployment and confirmation of complete occlusion of the patent ductus arteriosus via aortography; (F) Amplatzer Septal Occluder (white arrow) deployment in the intracardiac echocardiography. ADO, Amplatzer Duct Occluder; Ao, aorta; ASD, atrial septal defect; ASO, Amplatzer Septal Occluder; AV, aortic valve; ICE, intracardiac echocardiography; LA, left atrium; LV, left ventricle; PA, pulmonary artery; PDA, patent ductus arteriosus; RA, right atrium.

shunt through the ADO. As the thrombocytopenia remained for 3 months after the procedure, we had to discontinue antiplatelet therapy. Then, the platelet count recovered to the normal limits. Thrombocytopenia and haemolysis are known to be related to incomplete PDA closure.<sup>8</sup> The mechanism by which this occurs is believed to be the destruction of platelets and red blood cells by the residual shunt through the ADO. Moreover, according to a previous report, all patients who had haemolysis had residual shunts after PDA occlusion.<sup>8</sup> A study reported that the rate of complete closure of PDA by ADO at the time of the procedure was 89% for the paediatric patients.<sup>9</sup> Another study showed that haemolysis occurred in two of 114 paediatric patients who underwent transcatheter closure of PDA.<sup>10</sup> Additionally, antiplatelet therapy that prevents device-related thrombus formation after ASD closure can possibly inhibit the disappearance of a residual PDA shunt and exacerbate thrombocytopenia and haemolysis.<sup>11</sup> Another study reported that these complications followed by PDA closure occur less frequently in paediatric patients than in adult patients.<sup>12</sup> This is because, in part, a paediatric ductus arteriosus has less atherosclerotic changes. Moreover, its flexibility might enable the control of the shunt because

of good ADO properties. To prevent such serious complications, adult patients with PDA and ASD can benefit from PDA closure that precedes ASD closure.

In addition, the closure of PDA removes the volume overload of the LA and left ventricle, consequently leading to a decrease in the amount of the ASD shunt from the LA to the RA. On the other hand, if ASD closure precedes PDA closure, the volume overload in the RA improves. Nevertheless, blood flow through the PDA shunt would increase, resulting in a worsening of pulmonary congestion. The two-stage transcatheter closure, PDA closure that precedes ASD closure, can less drastically affect haemodynamics compared with the simultaneous combined closure.

We treated an adult patient complicating with both ASD and PDA and were able to safely achieve a successful outcome with two-stage transcatheter closure for ASD and PDA. From the perspectives of antiplatelet therapy and haemodynamic changes, it is reasonable that transcatheter closure for PDA should precede ASD closure. To the best of our knowledge, this is the first case report of such a staged treatment strategy in a patient complicating with both ASD and PDA.

## Lead author biography



Yusuke Soma entered Keio University School of Medicine in Tokyo in 2007. After graduation, he became a junior resident in Saitama City Hospital. Then, he entered the Department of Cardiology in Keio University Hospital in 2017. Currently, he is a graduate student of Keio University and is researching cardiac regenerative medicine.

## Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

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**Slide sets:** A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

**Consent:** The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

**Conflict of interest:** none declared.

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