CASE REPORT

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Multiple atrial septal defects: How and when should they be repaired? A case report

Yuli Sutarmini[1,3*], Ida Bagus Rangga Wibhuti[1], Ni Made Ayu Wulan Sari[1], Luh Oliva Saraswati Suastika[1], Satria Yudha Dewangga[2]

[1]Division of Echocardiography, Department of Cardiology and Vascular Medicine, Prof. Dr. I.G.N.G. Ngoerah General Hospital, Denpasar, Indonesia
[2]Division of Adult Congenital Heart Disease, Department of Cardiology and Vascular Medicine, Prof. Dr. I.G.N.G. Ngoerah General Hospital, Denpasar, Indonesia
[3]Cardiology and Vascular Medicine Department, Bali Mandara Public Hospital, Bali, Indonesia

*Corresponding author:
Yuli Sutarmini; Division of Echocardiography, Department of Cardiology and Vascular Medicine, Prof. Dr. I.G.N.G. Ngoerah General Hospital, Denpasar, Indonesia;
sang.medicine@yahoo.com

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ABSTRACT

Background: Atrial Septal Defect (ASD) is one of the most common congenital heart diseases. Percutaneous transcatheter closure (PTC) has become an alternative to surgery, especially in cases of secundum ASD. PTC in patients with secundum ASD with specific deficiencies, such as multiple rim deficiencies and multiple defects, remains a significant challenge. There is still considerable debate about whether a single device is sufficient to close two or more defects, or whether two or more devices are needed if the defects are not close to each other.

Case presentation: This case is a 41-year-old woman with multiple secundum ASDs and adequate rims where the large ASD (1st ASD) was successfully closed with a Lifetech Cera device with zero fluoroscopy, the insertion of which was guided by transesophageal echocardiography (TEE) and still left a small defect (2nd ASD) with several considerations.

Conclusion: A thorough assessment of the anatomy and hemodynamics of patients with multiple secundum ASDs is crucial. Closure of multiple secundum ASD defects using PTC depends on several factors, including the presence or absence of pulmonary hypertension, the number of defects, and their size. These factors can influence the decision to close multiple defects and are also crucial in selecting the appropriate device based on the size of the defect to be closed.

Keywords: adult congenital heart disease; multiple atrial septal defects; percutaneous closure of atrial septal defect; transesophageal echocardiography'

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INTRODUCTION

The incidence of congenital heart disease (CHD) varies around 4-50 / 1000 live births. At the global level, Atrial Septal Defect (ASD) is the 4th highest CHD, the incidence of ASD is estimated at 56 per 100,000 live births and 65-70% of ASD patients are estimated to have secundum ASD.[1] In Indonesia, the prevalence of CHD is quite significant, namely around 40,934 cases per year or 8-10 per 1000 live births with ASD contributing around 17% of the total cases, namely around 5,440 - 6,800 cases per year.[2,3]

If not treated properly, ASD can cause complications such as dilation of the RA and RV, pulmonary hypertension, arrhythmias and right heart failure. Clinical manifestations that can occur vary greatly depending on the size of the ASD and the direction of the flow or shunt present. Symptoms that often appear in adulthood are shortness of breath, palpitations, fatigue, and other signs of right heart failure.

There are two treatment options available for ASD patients, surgical techniques and PTC techniques. Surgical closure of ASD defects is generally safe, but is often associated with high morbidity, discomfort, significant scarring, and prolonged hospital stays. Percutaneous transcatheter closure (PTC) is an alternative, as it offers faster recovery, fewer complications, and no scarring.[4-6]

Echocardiography plays a crucial role in diagnosing ASD by assessing the defect size, morphology, and rims, as well as guiding and evaluating the results of ASD closure. Both TTE and TEE, especially with Three-dimensional (3D) imaging, are crucial for more accurate assessment and the presence or absence of crucial factors that determine the feasibility and success of PTC.[7,8]

In patients with multiple ASD defects, ASD closure with PTC may be more technically challenging and carries a higher risk of complications, such as device embolization, erosion, or residual shunt. Therefore, careful anatomical and hemodynamic assessment is necessary to determine the appropriate patient for PTC.[9]

CASE REPORT

A 41-year-old female patient was referred to the hospital with complaints of shortness of breath. Vital signs revealed blood pressure of 120/62 mmHg, pulse rate of 100x/minute, respiration rate of 20x/minute, and saturation of 98%. Physical examination revealed a grade 3/6 midsystolic murmur at intercostal space (ICS) II-III parasternal left line. There were no rhonchi or wheezing. There was no ascites or swelling in both extremities. Electrocardiography (ECG) revealed sinus tachycardia of 102x/minute, occasional premature ventricular contraction (PVC), P Mitral, right ventricular hypertrophy (RVH), right axis deviation (RAD). Chest X-ray revealed cardiomegaly with suspicion of pulmonary hypertension and left pleural effusion.

Transthoracic echocardiography (TTE) results with atrial situs solitus, large ASD secundum (diameter 20 mm) bidirectional shunt dominant L-R shunt, mitral rim 16 mm, posterior rim 25 mm, right atrium (RA) and right ventricle (RV) dilation, normal left ventricle (LV) and RV systolic function, severe tricuspid regurgitation (TR) with high probability of pulmonary hypertension (PH), moderate pulmonal regurgitation (PR), mild mitral regurgitation (MR).

Transesophageal echocardiography (TEE) results with multiple ASD secundum L-R shunt, floppy inter atrial septum (IAS; 1st ASD size with 3D measurement 18.3 mm x 10.7 mm and 2nd ASD size 6 mm x 3 mm, largest ASD diameter size two dimensional-2D 18 mm; **Figure 1**) posterior rim 12.27 mm, mitral rim 15.07 mm, SVC rim 17.11 mm, inferior vena cava (IVC) rim 14.64 mm, Aortic rim minimum 3.05 mm), the distance between the first and second ASD is 1.22 cm, mild MR, mild PR, moderate TR with high probability of PH, RA and RV dilatation, normal LV and RV systolic function (EF biplane-BP 69.4%, TAPSE 29 mm).

The second TEE evaluation was performed shortly before the ASD closure procedure (**Figure 2**) with the results showing multiple large, oval-shaped ASD secundum L-R shunts with a diameter of 21 mm x 11 mm for 1st ASD (antero-inferior) and 3 x 3 mm for 2nd ASD (supero-posterior) with adequate rims, so it was planned to perform transcatheter ASD closure with Cera ASD Lifetech No. 22 using the zero fluoroscopy method.

After administering anesthetic drugs, a 6F sheath was inserted through the femoral vein, then a 5F MPA2 catheter crossed into the left upper pulmonary vein (LUPV) then an extra-stiff Amplatzer wire was inserted into the LUPV and the 6F sheath was removed. Then a 12F Lifetech

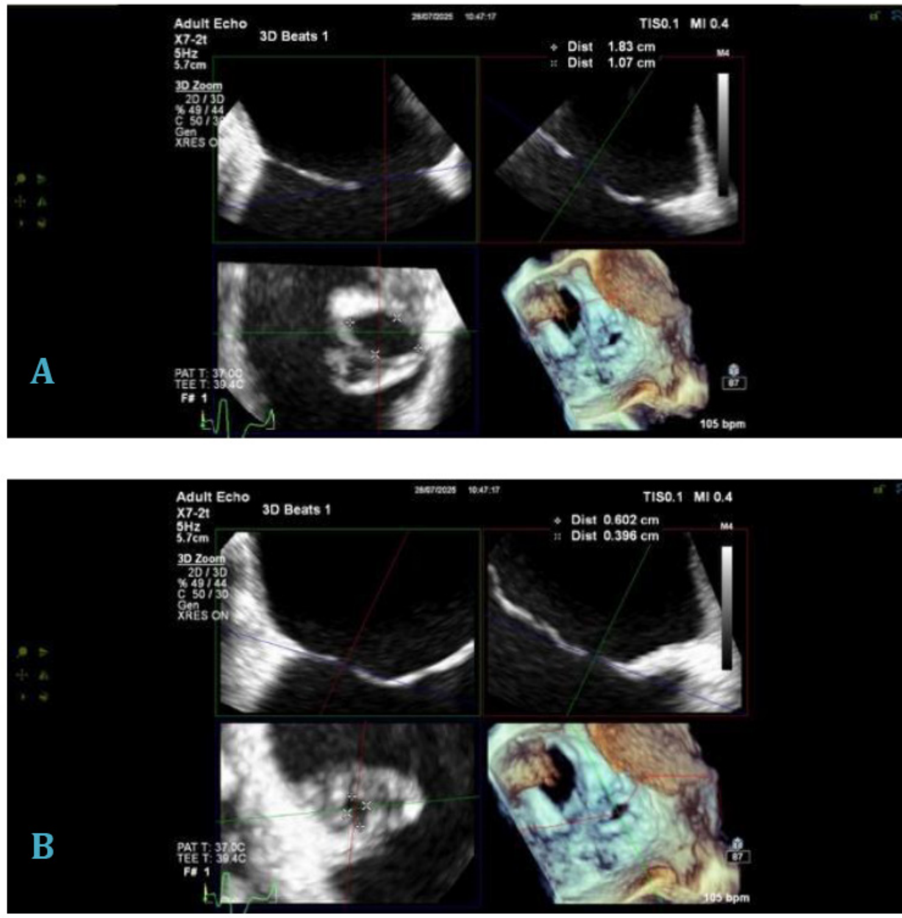

Figure 1. A. 3D size image on 1stASD; B. 3D size image on 2nd ASD

Figure 2. 3D size image on 1stASD

SFA delivery sheath was inserted into the LUPV, Real-time three-dimensional TEE was performed to assess whether the wire and sheath entered the defect hole to be closed (**Figure 3**). Then the Lifetech Cera ASD closure device no. 22 was inserted through the delivery sheath into the LUPV then the LA disk was opened in the LA and the RA disk was opened in the RA. After that, a TEE evaluation was carried out again along with a wiggle test and the device position was seen to be stable, no residual shunt was seen in 1st ASD, there was a residual shunt in the 2ndASD with left to right shunt (**Figure 4**). Next, the patient was scheduled for a TTE evaluation one month post-ASD closure and periodic evaluations were carried out to monitor the patient's condition where if the patient still had complaints related to the remaining defect (2ndASD) then ASD closure would be considered for 2ndASD.

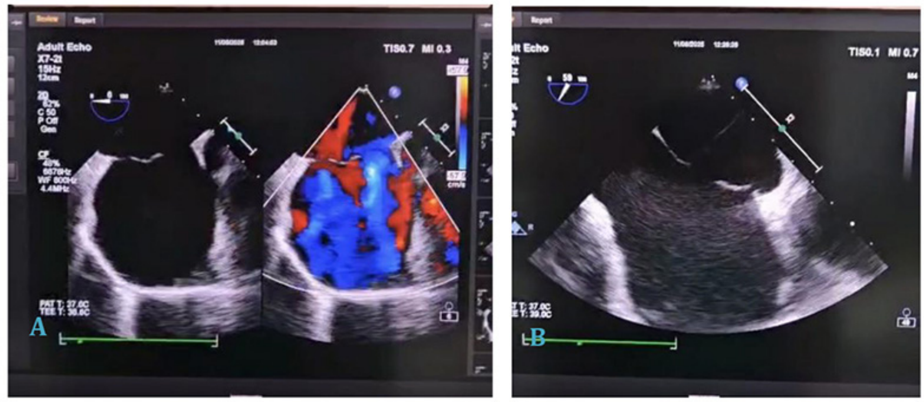

Figure 3. A. 2D color view from 1st ASD (anteroinferior), 2ndASD (supero-posterior) at 0 degrees; B. 2D view from 1st ASD (anteroinferior), 2ndASD (supero-posterior)

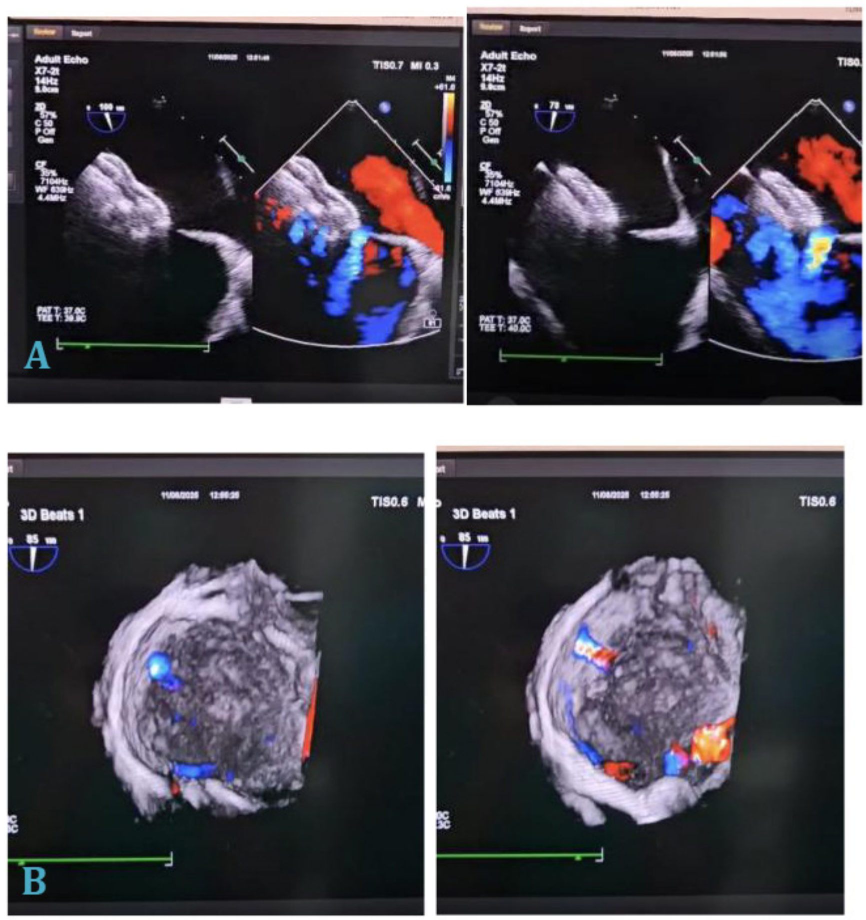

Figure 4. A. 2D color view showed device position was stable, no residual shunt on 1st ASD, and residual shunt on the 2nd ASD with left to right shunt; B. 3D view with residual shunt on the 2nd ASD with left to right shunt

DISCUSSION

Echocardiography is the primary examination for diagnosing ASD. TTE provides basic information on chamber size, defect size, ASD shunt direction, and pulmonary artery pressure. TEE, particularly three-dimensional (3D), provides more specific and comprehensive information, including defect size, number of defects, defect shape, and rim condition.[9,10] Adequate ASD rim coverage is crucial for PTC treatment of ASD. Rim deficiencies, particularly in the IVC, SVC, and posterior rims, can pose a high risk of device failure due to device instability, embolization, and residual shunt.[11-15]

ASD closure using PTC is an option if no other significant complications are found because it has a higher success rate and a lower risk of complications in both children and adults.[16-20]

ASD with multiple defects can be found in 10% of ASD patients.[20] Defect closure can be performed using more than one device if necessary, with good results.[21] However, in certain circumstances, complications can also arise, the most common being device embolization and erosion. Therefore, the anatomy of the ASD must be thoroughly assessed to determine whether there are complications such as rim deficiency or a floppy septum. In some cases, closure of ASDs with multiple defects can also be performed with a single device if the distance between the defects is relatively close (<0.7-1 cm), or if the device used has sufficient disc width to cover both defects. In multiple ASDs with relatively wide distances between the defects and severe rim deficiency, surgical closure of the ASD is recommended.[18,19]

Device measurement and selection are crucial for the success of the procedure. Device size selection depends on the size and shape of the defect. A wiggle test (Minnesota test) is also crucial to ensure device stability.[14,15]

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Balloon sizing is a method that can be performed before determining the device size, but as technology develops, at our center, imaging tools such as TEE and 3D echocardiography have become increasingly better in quality so that they can provide more precise images and measurements so that balloon sizing examinations are not carried out in this case, in addition to that, the procedure will be safer and more effective. Recent studies have shown that ASD closure can now be performed without balloon sizing and simply by relying on TEE results.[21-25] Awad et al. reported that selecting a device size 30-40% larger than the defect size yielded good results without complications.[22] Besides that, balloon sizing can sometimes overestimate the defect, leading to oversized devices, while imaging helps identify firm rims, allowing for more accurate device selection and potentially smaller devices. However, some operators still use this balloon sizing technique to identify other, previously unseen defects and to determine closure strategies. Furthermore, it has been suggested that without balloon sizing, PTC procedures result in shorter radiation times and a lower risk of oversizing.[26,27]

An ASD less than 1 cm (10 mm) usually does not need to be closed if it is asymptomatic and does not cause significant strain on the heart, so that without any complaints and with stable hemodynamics, the defect can still be observed first, but if the symptoms are significant and there are hemodynamic disturbances, the defect should be closed. Closure is generally recommended for defects that are considered hemodynamically significant, such as there is evidence of right heart enlargement or volume overload on an echocardiogram, the presence of symptoms such as shortness of breath, fatigue, or abnormal heart rhythms (arrhythmias), regardless of the defect size, the risk of long-term complications and a history of paradoxical embolism (a stroke or transient ischemic attack caused by a blood clot passing through the hole).[26]

Hemodynamically significant ASDs, typically defined as those with a Qp:Qs ratio greater than 1.5:1, should be closed to prevent long-term complications such as right heart enlargement, pulmonary hypertension, arrhythmias, and heart failure. Symptomatic patients, regardless of age, should undergo ASD closure to improve exercise tolerance and reduce symptoms. Asymptomatic patients with significant shunts (Qp:Qs ratio greater than 1.5:1) may also benefit from closure to prevent long-term complications, provided that systolic PA pressure is less than 50% of systolic systemic pressure and pulmonary vascular resistance is less than one third of the systemic vascular resistance.[27,28]

The clinical implications of this case show that with proper anatomical and hemodynamic evaluation, ASD closure with the PTC technique can be performed safely although it does not apply to certain patient conditions. Individual assessment remains important, taking into account echocardiography results, hemodynamic data, and patient-specific considerations.

CONCLUSION

PTC in patients with ASDs is a safe and effective procedure. Comprehensive echocardiographic assessment, particularly TEE and 3D, is essential to evaluate defect size, number, morphology, and rim adequacy. In cases of multiple secundum ASDs, PTC closure can still be performed, with various considerations depending on the number and size of the defects. Conservative options may be considered in cases of very small multiple secundum ASDs, in the absence of pulmonary hypertension, and with relatively normal heart function.

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ETHICS DECLARATIONS

Written and informed consent has been obtained from the patient to publish the case report.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTION

All authors have a proportional contribution to this study.

GENERATIVE AI DISCLOSURE STATEMENT

The authors declare that no generative Artificial Intelligence (AI) tools were used in the preparation, writing, data analysis, interpretation, or editing of this manuscript. The entire content of this article is the original work of the authors. The authors take full responsibility for the integrity and accuracy of the manuscript.

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