

Device Closure in Multiple Atrial Septal Defect Secundum Concomitant with Atrial Flutter

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Abstract:- This study discusses the case of a 23-year-old male with multiple atrial septal defect (ASD) secundum and atrial flutter who underwent ASD closure using device closure with satisfactory results. The patient recovered successfully and was discharged in good condition. ASD is a congenital heart disease often encountered in adulthood, with various types based on the location of the defect. Device closure of ASDs is one method to address abnormal blood flow, which can also reduce the risk of atrial arrhythmias through a negative remodeling process in the atria and ventricles. ASD closure in cases with multiple ASDs requires special consideration regarding the size, morphology, and distance between defects. Atrial flutter in ASD patients can be resolved with transcatheter ablation. Other studies have shown that device closure of ASDs is associated with a reduced prevalence of atrial tachyarrhythmia in the short to medium term, although atrial flutter/fibrillation and other arrhythmias are common in patients undergoing transcatheter ASD closure. Arrhythmias and conduction disturbances are also associated with ASD.

Keywords:- Atrial Septal Defect (ASD); Device Closure; Multiple ASD Secundum; Atrial Flutter; Transesophageal Echocardiography (TEE).

I. INTRODUCTION

Atrial Septal Defect (ASD) is a common congenital heart disease, with an estimated prevalence of 1.6 in 1000 births, and an estimated 97% of ASD patients survive to adulthood. Secondary ASD is the most common type of ASD, 75% of all ASD patients are secondary ASD and is more common in females than males with a ratio of 2:1. The diagnosis of ASD is often found when the patient is an adult. Based on the data, 10-15% of ASD patients have more than one hole defect or what is called Multiple ASD (Kuijpers et al, 2015). ASD closure using percutaneous devices has evolved and has been widely applied clinically with optimal results, but this technique is still difficult to apply especially in patients with large ASDs and in ASDs with complex morphology, due to the residual flow that is often found after the procedure. There are not many research reports that discuss multiple ASD closure techniques using a single device, especially in multiple ASD patients with various types and morphologies (Farhaj et al, 2019). Device closure of multiple atrial septal defects secundum concomitant with atrial flutter is a complex yet increasingly common procedure. The use of transcatheter

devices like the Amplatzer septal occluder has revolutionized the management of atrial septal defects (ASDs) (Gupta et al., 2011). These devices offer high success rates and low complication rates, making them a preferred choice for closure procedures (Gupta et al., 2011). Studies have shown that timely closure of ASDs can help reduce the morbidity associated with atrial flutter and fibrillation (Gatzoulis et al., 1999). The transcatheter closure of secundum ASDs has gained widespread acceptance due to its effectiveness and safety profile (Carano et al., 2001). The Amplatzer septal occluder, in particular, has been extensively used for closing secundum ASDs, providing successful outcomes in both children and adults (Carano et al., 2001; Demkow et al., 2001). The procedure's success often depends on the presence of adequate septal tissue surrounding the defect to ensure proper device placement and attachment (Hausdorf et al., 1996). In cases where patients present with both atrial septal defects and other cardiac issues like pulmonary valve stenosis, a combined approach of percutaneous pulmonary valvuloplasty and ASD closure has been successful in addressing the complex pathophysiology (Wahl et al., 2001). Additionally, the use of echocardiographic assessment has become instrumental in guiding percutaneous closure procedures, especially for multiple ASDs (Mitchell et al., 2004; Cao, 2000). While device closure of ASDs has shown excellent short and mid-term safety and efficacy, there are rare instances of complications such as device embolization or late infective endocarditis associated with occluder devices (Wei et al., 2012; Sanjuelo et al., 2022). However, these occurrences are infrequent and do not diminish the overall benefits of device closure procedures. In conclusion, the transcatheter closure of multiple atrial septal defects secundum concomitant with atrial flutter using devices like the Amplatzer septal occluder is a well-established and effective intervention. The procedure offers a minimally invasive approach with high success rates and low complication rates, making it a preferred choice for patients requiring ASD closure.

II. LITERATURE REVIEW

Device closure of multiple atrial septal defects secundum concomitant with atrial flutter presents a complex scenario. The Amplatzer septal occluder is a widely used device known for its high success rate and low complication incidence in closing atrial septal defects (Gupta et al., 2011). While centrally located secundum defects are considered ideal for device closure, there exists

significant morphological variation in the size and location of these defects, which can pose challenges during closure procedures (Mitchell et al., 2004). The transcatheter closure of atrial septal defects, including ostium secundum defects, has become a standard treatment due to its effectiveness and safety profile (Berger et al., 1999). However, the closure of multiple fenestrated secundum atrial septal defects remains a challenge in some cases, where surgical intervention may be necessary for effective closure (Carano et al., 2001). Successful device closure of atrial septal defects heavily relies on the precise placement of the closure device, ensuring that both the left and right atrial discs are appropriately positioned on either side of the atrial septum (Alobaidan et al., 2015). Understanding the detailed anatomy of atrial septal structures is crucial for the successful closure of defects (Pinto et al., 2007). The use of more than one Amplatzer septal occluder has been shown to be safe and effective in closing multiple atrial septal defects (Cao, 2000). Additionally, the outcomes of device closure for large and small secundum atrial septal defects have been compared, emphasizing the importance of rim anatomy in the closure process (Varma et al., 2003). In conclusion, device closure of multiple atrial septal defects secundum concomitant with atrial flutter requires careful consideration of the defect morphology, precise device placement, and an understanding of the anatomical structures involved. While the Amplatzer septal occluder is a commonly used device for such closures, challenges may arise in cases of multiple or complex defects, necessitating a multidisciplinary approach to ensure successful closure and optimal patient outcomes.

III. CASE PRESENTATION

A 23-year-old man was referred from East Luwu Hospital with complaints of shortness of breath and fatigue that had been felt since the last 4 years. The complaints were felt to be getting worse, especially since the last 1 year. The shortness of breath that appeared was felt to be worse during activity and decreased with rest. Shortness of breath is not worse when the patient is sleeping on his back. A history of waking up during sleep due to shortness of

breath was also denied. The patient also complained of chest palpitations that were felt in the last 3 months. Chest palpitations were felt to be intermittent, not influenced by activity. History of fainting was denied. The patient is the second child of 3 siblings, born full term assisted by a midwife, weight at birth is unknown. The patient's mother regularly visited the midwife during pregnancy, and there were no significant complications during pregnancy and childbirth. History of teratogenic drugs and herbal medicine consumption was denied. The patient had difficulty gaining weight as a toddler, and had recurrent respiratory infections. The patient had a history of Transesophageal Echocardiography (TEE) and Right Heart Catheterization (RHC) in March 2023 with the results of ASD bidirectional shunt and Pulmonary Hypertension High Flow Low Resistance, and was given sildenafil therapy and planned for RHC control and ASD closure 6 months later. The patient routinely visited a cardiologist at RSUD Luwu Timur and received routine therapy of warfarin 1 mg, concor 2 mg, and sildenafil 12.5 mg. The patient was then referred for RHC control and ASD closure if possible. From the results of the vital signs examination, it was found that the general condition appeared moderately ill, compos mentis consciousness, blood pressure 107/77 mmHg, heart rate 85 times per minute, breathing 18 times per minute, body temperature 36.6 degrees Celsius, and oxygen saturation 96%. Physical examination of the head and neck revealed non-anemic conjunctiva, non icteric sclera, JVP R+3 cm H₂O. Physical examination of the thorax revealed symmetrical static and dynamic chest development, equal tactile fremitus in both hemithorax, sonorous percussion in both hemithorax, bronchovesicular breath sounds, ronchi and wheezing were absent. Heart sounds S1 single and S2 normal split, irregular, and murmurs difficult to evaluate. Physical examination of the extremities showed warm acral and no pitting edema, cyanosis and clubbing finger. From the ECG results at PJT on August 29, 2023, an atrial flutter rhythm was obtained with variable conduction of 3:1, heart rate of 80 beats per minute, irregular, Right Axis Deviation, P wave sawtooth appearance, QRS duration of 0.12 seconds, crochete sign, and no ST segment and T wave changes.

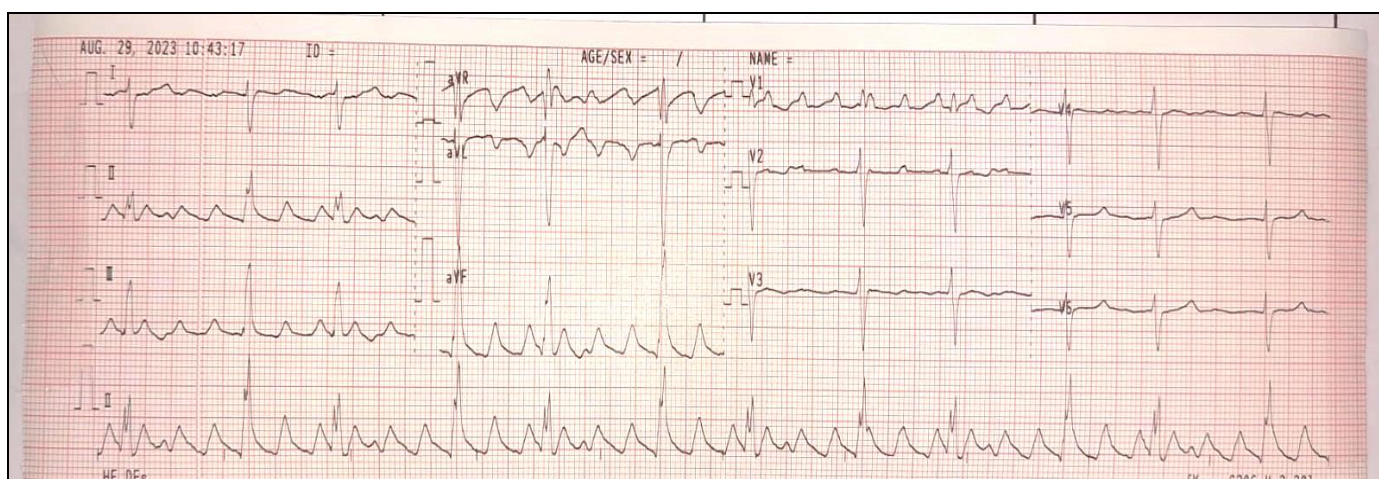


Fig 1: ECG of PJT RSWS August 29, 2023

(Atrial Flutter with Variable Conduction 3:1, HR 80 bpm, Irregularly Regular, RAD, Crochetage sign (+))

Echocardiography revealed dilated Left Atrium (LA), Right Atrium (RA), and Right Ventricle (RV) as well as D-shaped Left Ventricle (LV), and secundum ASD with 18-25 mm defect. Evaluation of the heart valves showed severe

tricuspid regurgitation, moderate mitral regurgitation, trivial pulmonary regurgitation, and decreased RV systolic function.

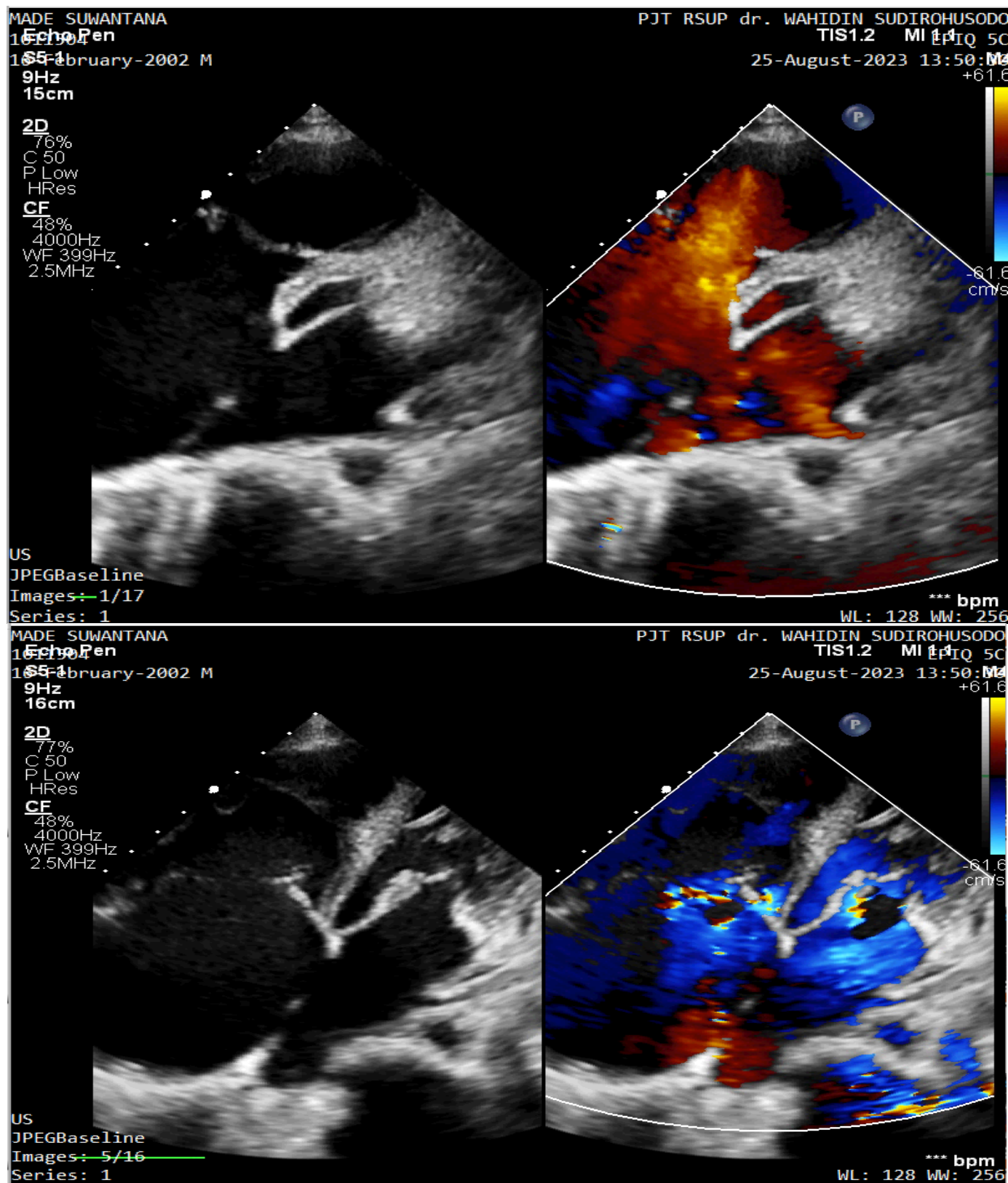


Fig 2: Transthoracic Echocardiography

Table 1: Results of Transtorakal Echocardiography

Echocardiography

- Site solitus
- AV-VA concordance
- All PVs drain into LA
- Cardiac chamber dimensions LA, RA, RV dilated, LV D-shaped
- IAS not intact, ASD secundum visible (defect size 18-25 mm, IAS 47-61 mm, mitral RIM 9 mm, Ao RIM 4 mm, posterior RIM 19-36 mm, SVC RIM 9 mm, IVC RIM 16 mm)
- IVS intact, no VSD seen
- Heart valves:
 - o Mitral: Moderate Mitral Regurgitation
 - o Aorta: 3 cusps, good function and movement
 - o Tricuspid: Severe Tricuspid Regurgitation (TR Vmax 4.13 m/s, TR Max PG 68 mmHg)
 - o Pulmonary: Trivial Pulmonary Regurgitation
- LV systolic function good, EF 76% (TEICH)
- RV systolic function decreased, TAPSE 1.4 cm
- PA confluence, no PDA seen
- No pericardial effusion seen
- Conclusion:
 - ASD Secundum Left to Right Shunt
 - Severe Tricuspid Regurgitation with High Probability of PH
 - Moderate Mitral Regurgitation
 - LA, RA, RV dilated
 - LV systolic function good, RV systolic function decreased

Based on the echocardiography results, the patient was planned for TEE (Transesophageal Echocardiography) and Right Heart Catheterization (RHC) or right heart catheterization evaluation and planned for ASD closure. From the TEE results, multiple ASD Left to Right shunts with diameters of 3 mm and 12.9 mm were found, with a suitable RIM size for closure using a device. From the right heart catheterization obtained pressure in the left atrium of 16 mmHg, pressure in the pulmonary artery of 70 mmHg, and pressure in the aorta of 91 mmHg, with oxygen saturation obtained saturation in the pulmonary artery of 85% and saturation in the aorta of 95%.

The patient was decided to close the ASD with a device with the help of guiding TEE. MPA2 6F catheter crossing from RA through ASD to LA, the catheter is then directed to PV and parked in LLPV. Next, an extrastiff wire 0.035 amplatzer was inserted through the catheter to the

LLPV. Then the MPA2 6F catheter and 6F sheath were removed, the amplatzer extrastiff wire was retained in the LLPV. Then Occlutech Delivery System 12F was inserted with the help of guiding the Amplatzer Exstrastiff wire to the LLPV, then the wire and dilator were removed. Occlutech Figulla Flex II ASD Occluder No. 36 was inserted through Occlutech Delivery System 12F with the help of guiding TEE and minimal fluoroscopy and placed in the LLPV through the ASD. Developed the first disk in LA, then the device was pulled up to the IAS and developed the second disk in RA, the disk was in good position. TEE and TTE evaluations were performed, the device appeared to be in good position, there was no residual shunt, and it was decided to release the ASD occluder device. From the evaluation results through TTE and TEE, it shows that the device position is good, and there is no residual shunt.

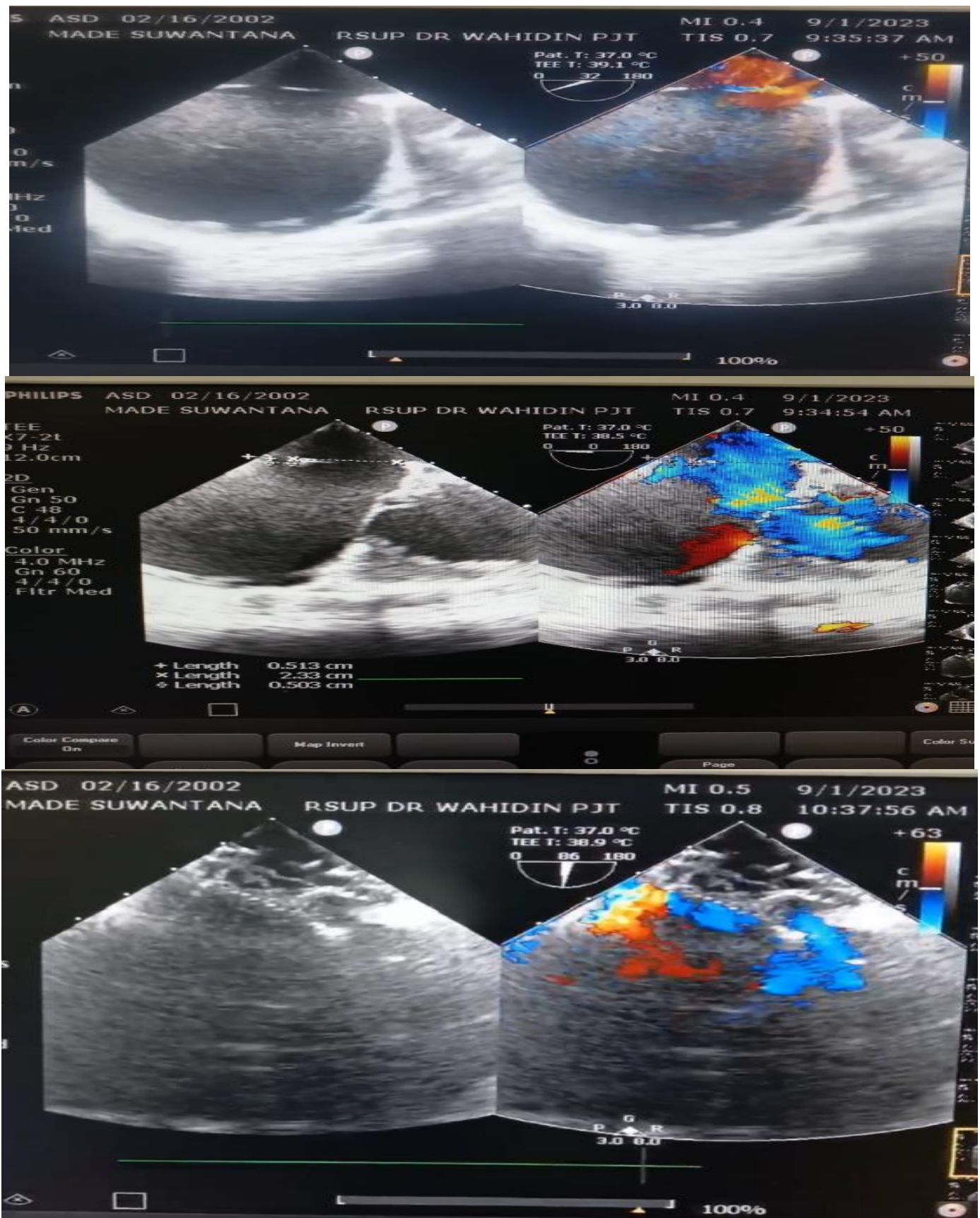


Fig 3: Transesophageal Echocardiography (TEE) and TEE Guiding Device Closure

TTE evaluation 6 hours post-action found the device well seated, and there was no residual shunt. A post-action control ECG revealed atrial flutter with variable

conduction. The patient was admitted to the treatment room and discharged 2 days post-action in good condition.



Fig 4: TTE Evaluation 6 Hours After Action

IV. DISCUSSION

A. Natural History ASD

Atrial septal defect (ASD) is the most common congenital heart disease in adults. The prevalence of ASD is estimated to be around 0.88 cases out of 1000 patients. The diagnosis of ASD is often made in the fourth decade of life, as almost all ASD patients are asymptomatic until the age of 40, at which age increased pulmonary blood flow can cause vascular remodeling and altered shunt flow, leading to decreased organ perfusion (Bradley et al, 2020). There are 4 types of ASD based on the location of the defect, namely ASD primum, which is a defect that arises due to the absence of tissue formation around the atrioventricular valve. ASD primum is associated with a gap on the left side of the atrioventricular valve and is often referred to as partial AVSD. The incidence of ASD primum is 10% of the ASD population. The most common type of ASD is ASD secundum, which accounts for about 80% of all ASD patients. ASD secundum results from a defect in the ostium secundum, which is a tissue deficiency in the atrium at the level of the fossa ovalis. Another type of ASD is a defect in the superior and inferior sinus venosus in the embryologic phase of the sinus venosus and is often accompanied by a partial anomaly of pulmonary venous

return, particularly in the right upper pulmonary vein. Another rare type of ASD is coronary sinus defect type ASD, also known as unroofed coronary sinus. Coronary sinus defect type ASD is usually difficult to find using traditional radiology modalities. This type of ASD can be detected by the bubble test, which looks for bubbles of saline fluid from the left upper extremity crossing into the left atrium before entering the right atrium (Bradley et al, 2020). Based on the physical examination and supporting examination, this patient's ASD can be confirmed as secundum ASD. In some adult ASD secundum patients, the increased circulating blood flow in the lungs due to ASD can form abnormal remodeling of the pulmonary vasculature, and increase pulmonary vascular resistance. This increased pulmonary vascular resistance can then lead to pulmonary arterial hypertension (PAH). In adult ASD patients, 16% of them experience PAH. Despite having PAH, in most cases, the pressure is still lower than the systemic blood pressure, but in a small number of cases, the pulmonary pressure can equal or exceed the systemic pressure, causing a change in flow or shunt from left to right (left to right shunt) to right to left (right to left shunt). This change in flow to right to left then causes peripheral desaturation accompanied by cyanosis, this condition is called Eisenmenger syndrome (Bradley et al, 2020).

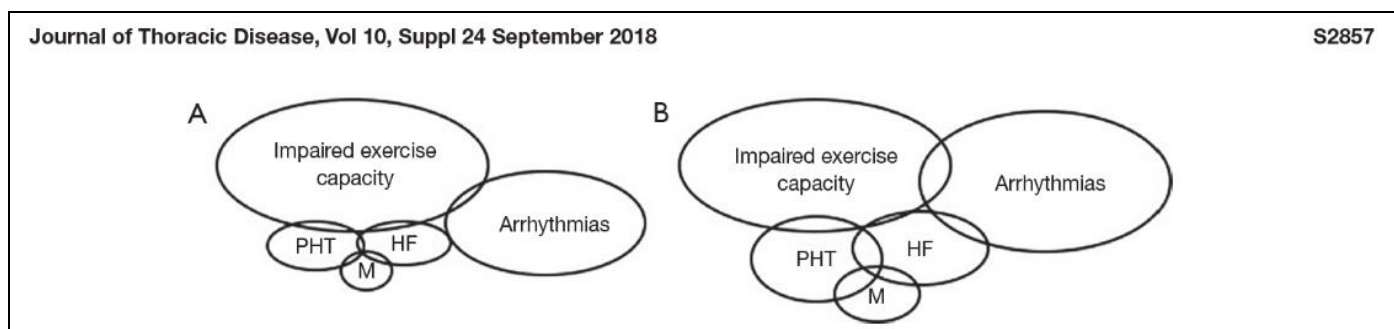


Fig 5: ASD4 Natural History of Patients with unoperated significant atrial septal defects, according to the age at evaluation: before 40 years old (A) and after 40 years old (B). The relative size of the circles represent the relative proportion of occurrence of cardiac adverse events. HF, heart failure; PHT, pulmonary hypertension; M, mortality
 Source: Le Gloan, 2018

In general, therapeutic options in Eisenmenger syndrome patients are very limited compared to uncorrected ASD patients with mild to moderate PAH. A number of studies have shown that ASD patients with mild to moderate PAH can respond well to PAH-specific therapy, and can potentially reduce pulmonary vascular pressure to a level where closure or correction can be performed, thereby improving patient morbidity and mortality (Bradley et al, 2020). In this patient, it was decided to close the ASD using a device after 6 months of PAH-specific therapy.

B. Device Closure in Multiple ASD Patients

Based on the size, morphology and distance between the defects, multipel ASD or Multi hole ASD (MHASD) is classified into 4 types. This classification aims to make it easier to choose the closure technique to be performed on the patient. Based on defect size, defect diameters >15 mm are classified as large defects, 5-15 mm are categorized as medium, and <5 mm are classified as small defects. The interdefects distance (IDD) is considered to be the distance between the center of the defect (the main defect where the device is implanted) and the surrounding defect hole to be covered (Farhaj et al, 2019).

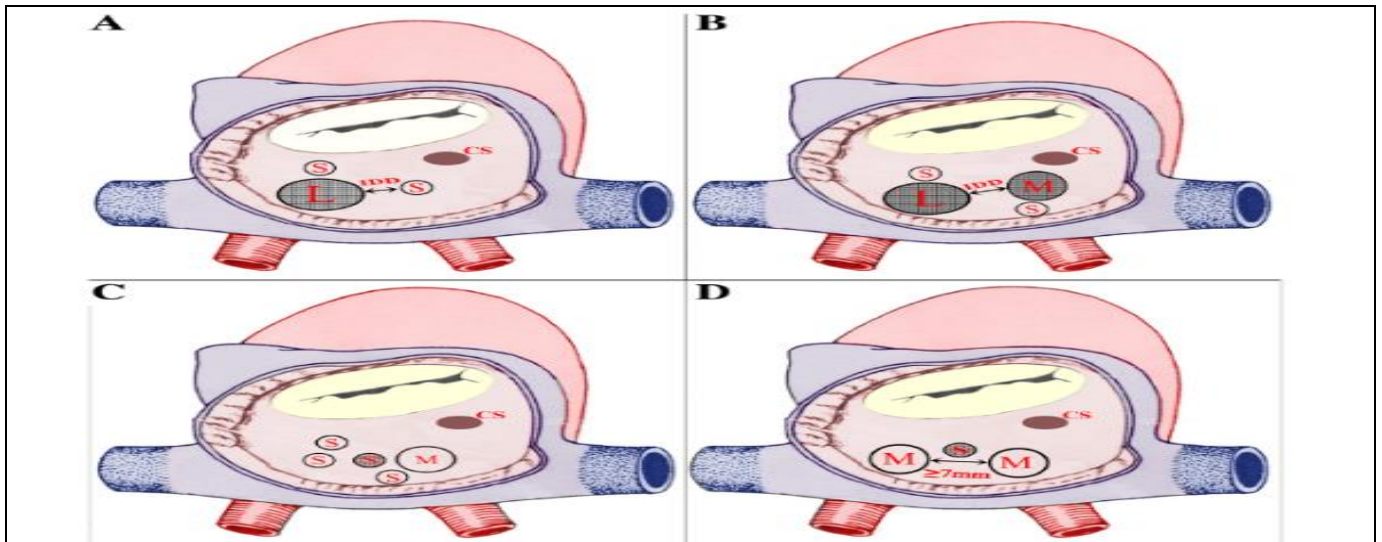


Fig 6: Multiple ASD Classification of Multi-Hole Atrial Septal Defects. (A) Type A, Large-Small Hole Configuration Accompanying Small Defect With Different IDD. (B) Type B, Large-Moderate Configuration Accompanying Small Defect with Different IDD. (C) Type C, Cribriform Formation, with the Defect Number of ≥ 5 . (D) Type D, Moderate-Small-Moderate Morphology. $IDD \geq 7$ mm. (L= Large; S= Small; M= Moderate; IDD =Inter-Defects Distance; Shadow

Device selection is done by considering various factors including the configuration of the defect to be closed, device implantation position, intracardiac structures around the defect, and defect size. In patients with type A and type B MHASD morphology, a single device atrial

septal occluder can generally be selected, but if the configuration is small and medium or both defects are small, a PF device can be considered. In type C and D MHASD, a PF device is recommended (Farhaj et al, 2019).

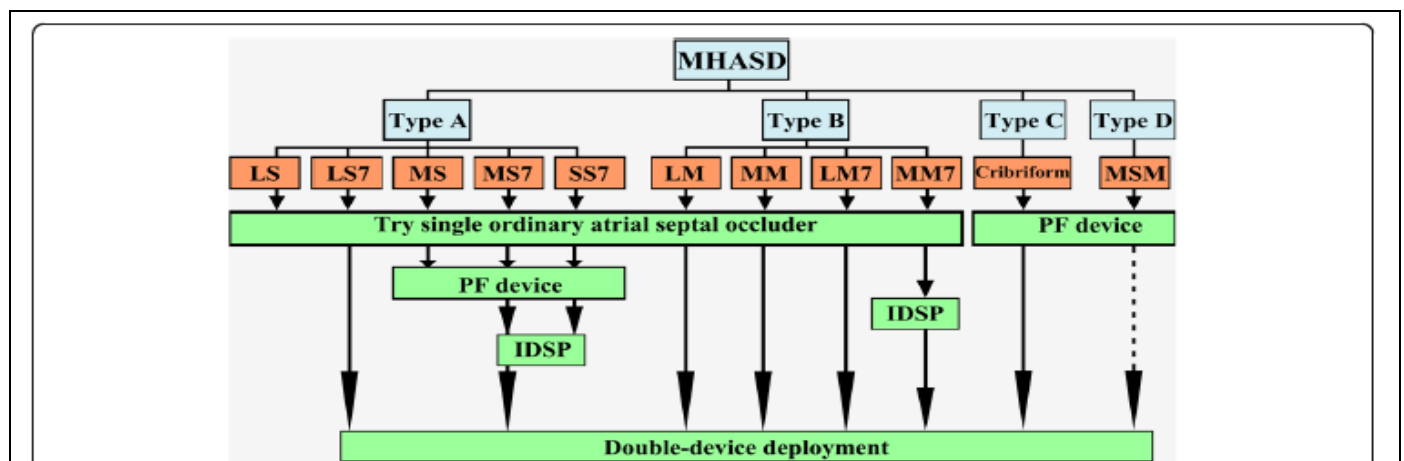


Fig 7: Multiple ASD Device Selection Flow Chart. Device Selection Flow Chart (LS = Large-Small; MS = Moderate-Small; SS = Small-Small; LM= Large Moderate; MM= Moderate- Moderate; MSM= Moderate-Small- Moderate Hole Configuration, IDSP = Inter-Defects Septal Puncture. The Numeric "7" Denotes Inter-Defects Distance of ≥ 7 mm.) Dotted arrowhead Line denotes Possibility of Double Device Deployment

A number of techniques and modalities can be used to evaluate multiple ASDs before and after closure, including Real Time 3-dimensional echocardiography (RT3D). The use of RT3D aims to confirm the anatomical location of the defect, determine the tissue structure around the defect and rim, and provide extensive visualization of the septum and the relationship and distance between ASD defects (Jung et al, 2018). Temporary Balloon Occlusion Test can also help to determine the compliance of the rim and septum around the defect so as to predict the possibility of rim and device changes after implantation. Fluoroscopic evaluation with balloon sizing can provide additional information regarding the spatial distance between the defect and septum. Small ASDs that are located adjacent to ASDs with large defects (distance between defects <7 mm) can be directly closed using a single device implanted in the large defect.⁸ This single device closure technique was applied in this case using Occlutech Figulla Flex II ASD Occluder no 36.

C. Atrial Arrhythmia in ASD

In addition to the presence of multiple hole ASD, this patient was also found to have atrial flutter. Atrial flutter and atrial fibrillation are advanced complications that are often found in ASD patients both before and after surgery. The prevalence of atrial arrhythmias in unoperated ASD patients increases significantly with age, especially in patients over the age of 40 years. Although a number of

studies have reported improvement in atrial arrhythmias in patients who underwent surgical or device closure, atrial flutter and atrial fibrillation remain short-term complications in a number of patients, approximately 10-25% of patients. Early ASD closure, especially in large ASDs and multi-hole ASDs, should be done to prevent worsening arrhythmias (Chiu et al, 2017). The process of atrial flutter in ASD patients is thought to occur due to 2 things, namely geometric remodeling and electrical remodeling. In geometric remodeling, left-to-right shunts can result in increased pressure and load in the right atrium resulting in stretching of the atrial wall. This process then continues until the atrial wall thickens and undergoes fibrosis. This is the substrate for atrial flutter and atrial fibrillation in ASD patients (Contractor et al, 2017). In addition to geometric remodeling, another pathomechanism that is thought to influence the occurrence of arrhythmias is electrical remodeling. This process includes increased recovery time at the sinus node, prolongation of intra atrial conduction, and increased effective refractory period of the atria. These factors predispose not only to bradyarrhythmias, but also to reentrant atrial arrhythmias. ASD not only blocks conduction but also causes the conduction circuit to rotate around the defect, which can lead to peri ASD flutter, even in the absence of previous surgery or device closure (Contractor et al, 2017).

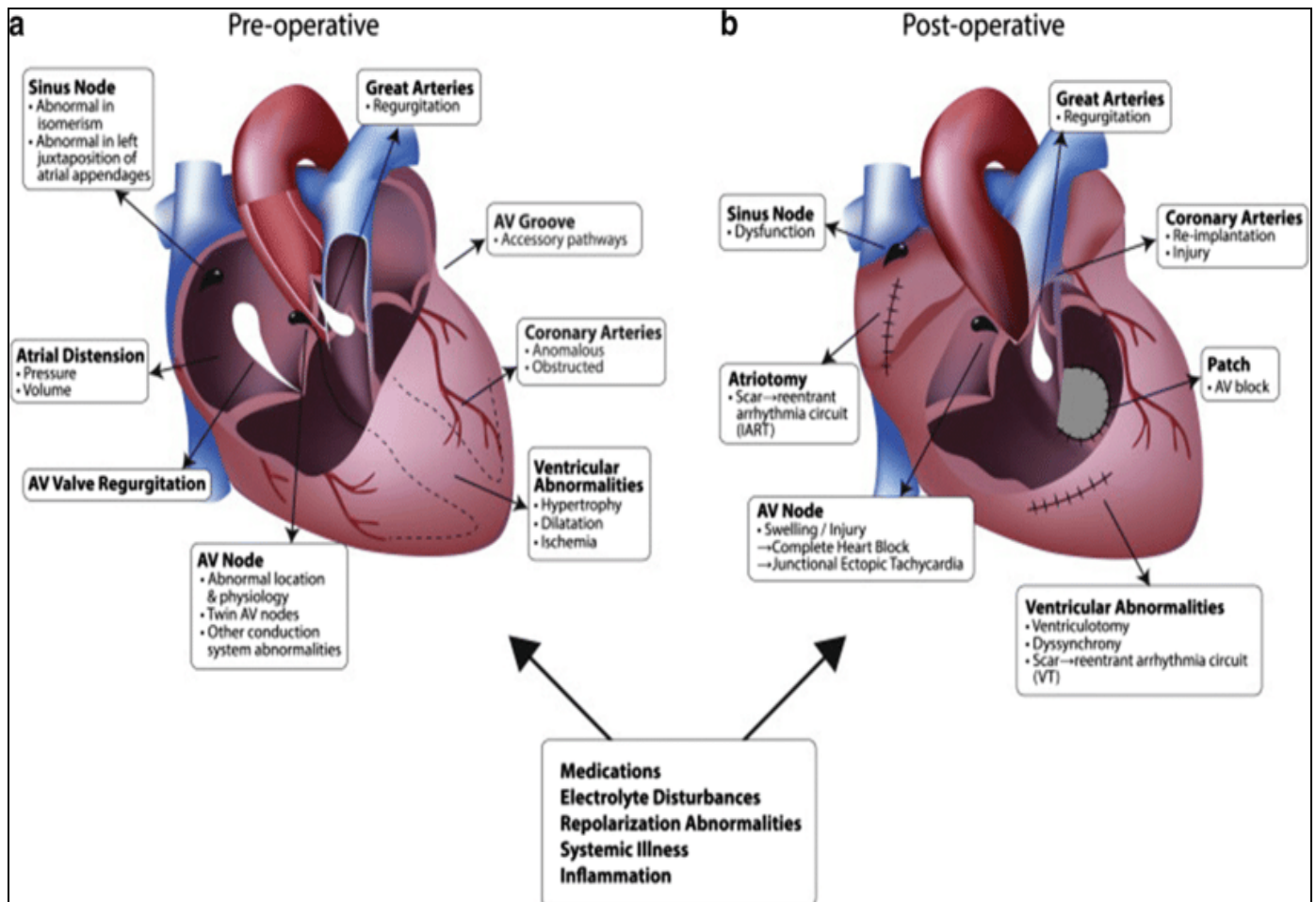


Fig 8: Factors That Trigger Arrhythmia in Congenital Heart Disease Patients

AVNRT is a frequent arrhythmia in patients with ASD, the diagnosis can be made by looking at P wave morphology, AV dissociation, and by performing several maneuvers. The most frequent atrial arrhythmias in ASD patients are CTI-dependent atrial flutter, atypical atrial reentrant tachycardia, atrial fibrillation and rarely focal atrial tachycardia. Although rare, it is important to rule out the presence of focal atrial tachycardia, given the different ablation targets between focal atrial tachycardia and macro reentrant circuits (Contractor et al, 2017). Defect closure in ASD may decrease the risk of arrhythmia through a negative process of remodeling in the right atrium and ventricle. The decrease in volume in the right atrium and ventricle can increase the right ventricular ejection fraction, and indirectly increase the left ventricular volume. Changes in hemodynamics and structural improvements also contribute to the reversal of electrical remodeling, which explains the anti-arrhythmic effect in ASD patients who have been closed (Vecht, 2010). In this patient, the atrial flutter that appeared was suspected to be a typical cavotricuspid isthmus-dependent atrial flutter that occurred in the double loop complex with reentry in the intra atrial circuit, where transcatheter ablation could be performed as a therapeutic measure (Williams & Perry, 2018).

V. CONCLUSION

Based on the information provided in this case report, atrial septal defect (ASD) closure using device closure in a 23-year-old male patient with multiple secundum ASDs and atrial flutter gave good results, with the patient recovering and being discharged in good condition. ASD is a congenital heart disease that is often found in adulthood, and ASD closure with a device is one method to overcome abnormal blood flow problems and reduce the risk of atrial arrhythmias through the process of negative remodeling in the atria and ventricles. ASD closure in cases with multiple ASDs requires special consideration regarding the size, morphology, and distance between defects. Atrial flutter in ASD patients can be resolved with transcatheter ablation. Other studies have shown that device closure of ASDs is associated with a reduced prevalence of atrial tachyarrhythmia in the short to medium term, although atrial flutter/fibrillation and other arrhythmias are common in patients undergoing transcatheter ASD closure. Arrhythmias and conduction disturbances are also associated with ASD.

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