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CASE REPORT

Fenestrated Atrial Septal Defect Percutaneously Occluded by a Single Device: Procedural and Financial Considerations

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ABSTRACT

A 45-year-old patient presented with a cerebrovascular attack and was subsequently found to have a multi-fenestrated atrial septal defect. Various therapeutic options for percutaneous transcatheter closure with their respective benefits and flaws are discussed, as well as procedural and financial considerations. The decision making process leading to a successful result using a single occlusive device is presented, alongside a review of the literature.

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INTRODUCTION

Atrial septal defects (ASDs) account for 10–17% of congenital cardiac anomalies [1]. Percutaneous closure of ostium secundum ASD is a safe and effective alternative to surgery [2]. Nearly 10% of patients with secundum-type ASD are found to have multi-fenestrated ASDs (mfASDs) [3]. The closure of more than one defect poses several challenges to the cardiologist. The authors describe a case of a woman with multiple ASDs who was successfully treated with a single ASD occluder.

CASE REPORT

The patient was a 45-year-old female who had a cerebrovascular attack (CVA), presenting with dysarthria and right hemiparesis. Computed tomography of the brain was interpreted as normal on the second day of symptoms. The patient's neurologic symptoms gradually

improved and the patient resumed her work as a school teacher 3 months following the acute event.

There was no history of smoking, the patient's blood pressure was normal, and blood tests for renal and liver functions were normal. Fasting blood glucose and cholesterol were within the normal limits. The patient underwent a Doppler study of carotid and vertebrobasilary arteries, which were found to be normal. Hypercoagulability workup was also normal.

The patient did not report palpitations. The patient's parents were treated for arterial hypertension. A 24-h electrocardiogram Holter study was normal, with no paroxysmal atrial fibrillation events and no other ectopic activity.

During a routine transthoracic echocardiogram (TTE) study, the right ventricle was noted to be mildly dilated and a small-to-moderate, left-to-right flow was observed across a fenestrated interatrial septum (IAS). There was normal biventricular function with no hypertrophy. Very mild, nonrheumatic mitral valve regurgitation was observed and minimal physiologic regurgitation of the tricuspid valve was noted with a systolic continuous wave Doppler gradient of 22 mmHg, resulting in a normal estimate of pulmonary artery pressure.

After informed consent was obtained, the patient underwent general anesthesia with endotracheal intubation. A transesophageal echocardiogram (TEE), performed in the catheterization laboratory for the guidance of transcatheter closure of the defect, revealed a floppy IAS with four fenestrations (Fig. 1). Agitated saline injection through the right femoral vein with simulation of Valsalva maneuver by the anesthesiologist induced a large amount of microbubbles shunting right-to-left across the IAS. A decision was made to close the defects using a single device. The authors used the Occlutech Figula® Flex ASD 15 mm (H. + H. Maslanka GmbH, Stockacker, Germany) occluder for this fenestrated IAS. The author sized and deployed the device in one of the central defects, which measured 15 mm. A leak still existed outside the perimeter of the device (Fig. 2). A second attempt was subsequently performed in the adjacent central defect, using the Occlutech Figula®

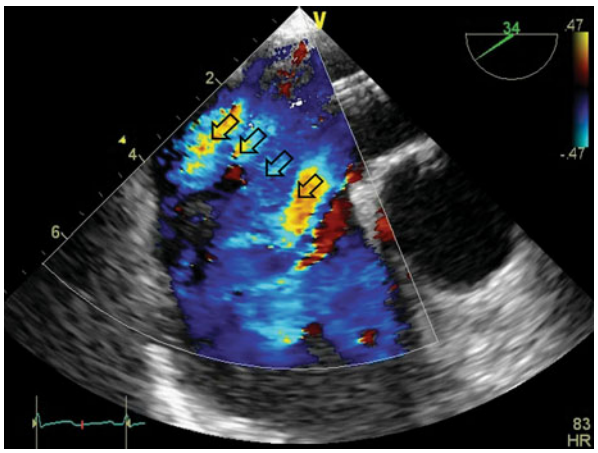

Fig. 1 A transesophageal echocardiogram demonstrating a multifenestrated atrial septal defect with four openings (*arrows*)

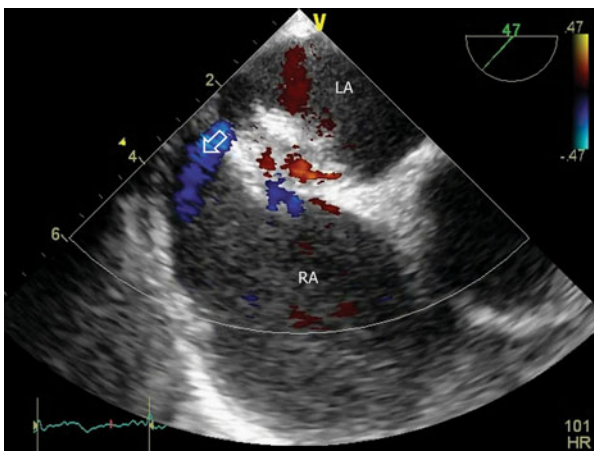

Fig. 2 A transesophageal echocardiogram demonstrating a deployed occluding device in one of the atrial septal defects. There is a leak outside the perimeter of the device (*arrow*). *LA* left atrium, *RA* right atrium

Flex ASD 21 mm (H. + H. Maslanka GmbH, Stockacker, Germany) occluder. Residual leak was demonstrated only within the perimeter of the device (Fig. 3). At that stage the device was released. The adequacy of device position was confirmed by gentle tagging of the deployed device prior to release and by TEE imaging. The procedure was uneventful and lasted 65 min, most of which was dedicated to thorough TEE investigation and planning. The screening time was 18 min.

During the procedure, the patient was given 6,000 units of heparin and 1,000 mg of cefazoline intravenously, followed by two additional cefazoline doses over 24 h.

Following the procedure, the patient was started on clopidogrel 75 mg/day for 3 months and aspirin 100 mg/day for 1 year to allow complete endothelialization of the occlusive device.

On a follow-up visit 1 month after the procedure, the device was found to be properly placed without any IAS leaks, confirmed via TEE. There was no interference with cardiac inflows and no impingement on heart valves. Currently, 6 months after the procedure, the patient is asymptomatic with no additional neurological events, and is leading a normal life.

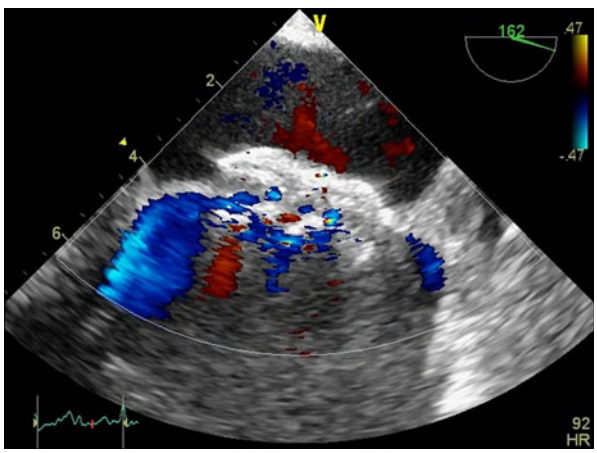

Fig. 3 A transesophageal echocardiogram demonstrating a deployed occluding device in one of the atrial septal defects. Residual shunt is present within the perimeter of the device (*arrow*)

DISCUSSION

Children and adults with secundum-type ASD may present with mfASDs. Clinical indications for closure of the defects are as for a single ASD, including symptomatic patients and/or increased pulmonary blood flow [pulmonary-to-systemic flow ratio (Q_p:Q_s) >1.5:1]. While surgery was the only available treatment for secundum ASD in the past, nowadays the preferable therapeutic modality is by the percutaneous approach. Although the transcatheter approach becomes the treatment of choice for ASD patients, surgical results remain the gold standard [4]. Surgery continues to be indicated for patients who prefer to avoid implantation of a foreign body, for defects that are too large to be closed by an implantable device or lack adequate rim for a device to lean on, and for unfavorable IAS morphology precluding successful and safe closure of the defects using one or several occlusive devices. In elderly patients, the prompt recovery following transcatheter ASD closure makes this alternative approach to surgery more attractive in view of prolonged convalescence and an increased rate of significant complications in the older population due to comorbidities [4].

Since the first reported transcatheter closure of secundum ASD by King et al. in 1976 [5], its use has been widely accepted as a highly effective and safe alternative to surgery [5]. The percutaneous transcatheter closure of fenestrated ASDs may be more complicated than closure of a single ASD, and requires

careful investigation of the anatomy of the defects and their surroundings, as well as prudent selection of the most appropriate method. The transcatheter closure of a multiple or fenestrated ASD can be accomplished by several methods.

The defects may be closed by the use of several devices, each of the devices implanted to close one or more defects. When the distance between the ASDs is greater than 7 mm, placement of two devices may be recommended [6]. Awad et al. [6] published a series of 33 patients who underwent multiple ASD closure using more than one device, with a 97% immediate success rate. There were two cases of complications. The first was device embolization and the second was device erosion due to oversizing of a device. The second case needed a surgical removal of the devices. The explanted devices showed complete endothelialization, refuting previous findings of noncovering due to devices overlap [7].

Closure of the larger defect should be performed first [6, 8]. The second device may have to be larger in order to overlap the rim of the first device, in spite of the smaller stretched diameter of the defect. The cardiologist may mistake a star-shaped defect for multiple small defects. This might cause the first smaller device to be easily pulled through upon deployment.

Awad et al. used the Amplatzer® septal occluder (AGA Medical Corporation, Plymouth, MN, USA) for all but one (who received two Amplatzer® Cribriform [AGA Medical Corporation, Plymouth, MN, USA] devices) of their series [6]. Other investigators have used Amplatzer® PFO occluders (AGA Medical Corporation, Plymouth, MN, USA), CardioSEAL® and STARflex® occluders (NMT Medical Inc., Boston, MA, USA), and GORE HELEX® septal occluders (W. L. Gore & Associates, Elkton, MD, USA) [8].

When using more than one device, attention should be paid to adequate distance from structures like the vena cavae entrances and the coronary sinus. The devices might interfere with blood flow and even increase the risk for thrombosis. This, however, has not been apparent in follow-up studies, even after cessation of antiplatelets therapy [9]. In addition, the devices might cause erosion of important tissues, including the aortic root, the atrioventricular valves, or the atrial free walls. Nevertheless, closure of multiple ASDs using multiple occluders seems to be a safe and effective method.

An additional financial issue that should be considered when implanting more than one device is the reimbursement system used by current health maintenance organizations (HMOs). Since the cost of percutaneous closure of ASD is reimbursed according to a specific diagnosis-related group (DRG), closing multiple ASDs using more than one device during a single procedure will exceed the DRG budget.

When multiple ASDs cannot be closed with one large device or two to three smaller devices, performing an atrial balloon septostomy followed by device closure of the single iatrogenic ASD has been proposed [10]. While it has been successfully accomplished previously, this method carries the risk of creating an unpredictable, irregular, large hole. Such a hole might not be readily closed percutaneously.

When the defects are in close proximity, an attempt may be made to close them all using a single device. Szkutnik et al. [11] reported its feasibility in 2004. A distance of less than 7 mm between the defects is considered appropriate for this procedure. A larger device should be employed in order to cover all the defects. However, it was suggested that even if residual leaks were observed they tend to resolve with

time [11]. In addition to the diameter of the device to be used, a decision has to be made regarding the type of device. A single regular ASD occluder inserted through the central or largest defect will be stabilized in place by its waist. The waist will also stretch the IAS; thus, bringing the surrounding defects in proximity and decreasing their size.

A fenestrated ASD is often associated with atrial septal aneurysm. Some cardiologists prefer to close these lesions using Cribriform devices [9, 12]. These are characterized by large discs and a narrow waist (or connecting pin). This structure means that the device does not rely on the flexible septum to stabilize its position but rather uses its larger discs to stabilize the aneurysmal septum. Straightening the aneurysmal septum may also decrease the size of the defects. The size of the device is determined by the "steady rim," which is the area that should include all defects [12]. Nevertheless, using a device which has an interconnecting pin rather than the regular waist means that after the release the device may cruise around the hole into which it was released, if the discs are not adherent to the IAS. Therefore, the final position may vary and should be re-inspected to ensure a proper defects closure.

The benefits of using a single device are a shorter procedure duration and lesser chance of interference with venous blood flow, atrioventricular valves function, or adjacent tissue erosion. However, an oversized device on a floppy IAS might cause tissue dissection or arrhythmogenicity. In the authors' experience of 541 ASD closures, 13% of which were fenestrated, only one case needed two devices for closure (unpublished data).

From the authors' experience, it is possible and advantageous to use a single device for mfASD closure. In comparison with the recently published data of the H. Sievert group [13], who reported 35.8% of patients with mfASD who were treated with more than one device, the authors' group implanted two devices in a single patient, which accounts for only 0.2% (unpublished data). The report staged procedure in 49 of the 53 patients who required multiple devices, which meant separate interventions for most patients who received more than one device [13]. The only patient that the authors treated with two devices had them implanted during one procedure. These data reflect a difference in the clinical approach rather than diversity in technical/procedural experience.

The present patient experienced a CVA at a relatively young age. A thorough investigation did not disclose any apparent etiology for this event other than a cryptogenic stroke due to a paradoxical embolus through the mfASD. Although the patient's interatrial shunt was left-to-right, events such as Valsalva maneuver and cough may transiently reverse the flow, leading to paradoxical emboli and CVA. Furthermore, these defects caused right ventricular enlargement with pulmonary overflow. These findings and the patient's CVA history were indicative for ASD closure. The expected benefits for the present patient included prevention of a recurrent stroke due to a presumed paradoxical emboli and prevention of further cardiac complications including arrhythmia, heart failure, and deteriorating functional capacity. There was also a risk (though relatively low) for increasing pulmonary vascular resistance without ASD closure.

After an informed consent was obtained from the patient, a TEE was performed in the cardiac catheterization laboratory. It demonstrated a floppy IAS and a mfASD with four openings in close proximity. The exact distance between the defects could not be measured as they were in different anatomical planes. The authors decided to close the mfASD using a single device.

CONCLUSION

The authors described the management of a patient with multiple ASDs, discussed the different therapeutic options for percutaneous transcatheter closure with their respective benefits and flaws, and described the decision-making process leading to the successful result. Future trials should compare the complications and success rate of mfASD closure using a single device versus multiple devices, and single versus staged procedures. It should be considered whether a surgical approach may be preferable in complex cases of mfASDs requiring multiple devices.

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