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CASE SERIES

Device Closure of Superior Sinus Venosus Atrial Septal Defects

A single centre experience

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ABSTRACT: Sinus venosus atrial septal defects present a wide variety of anatomical features and are frequently associated with partial anomalous pulmonary venous drainage of one or more right pulmonary veins. Surgical correction used to be the standard treatment. In recent times, transcatheter correction of superior sinus venosus atrial septal defects has come into vogue. The transcatheter closure of these defects with covered stents at a tertiary care centre in Oman between 2018 and 2023 is reported.

Keywords: Heart Septal Defects, Atrial; Vena Cava, Superior; Endovascular Procedures; Stents; Cardiac Catheterization; Echocardiography, Transesophageal; Oman.

Sinus venosus atrial septal defects (SVASDs) represent approximately 4–11% of all atrial septal defects.[1] The current report mentions the superior SVASDs that are frequently associated with partial anomalous pulmonary venous drainage of the right upper and/or right middle pulmonary veins. The drainage of the right upper pulmonary vein (RUPV) is into the right atrium at the mouth of the superior vena cava (SVC) and is caused by a deficiency in the in-folding of the atrial wall which forms the posterior wall of the SVC and the anterior wall of the RUPV.[2] This frequent coexistence of anomalous pulmonary venous drainage in SVASDs contributes to an additional left-to-right shunt resulting in volume overload of the right ventricle and possible pulmonary hypertension. The development of Eisenmenger syndrome may occur earlier in these patients compared to those with an isolated atrial septal defect.[3] Hence closure of these defects once they are detected is indicated. We report a series of transcatheter closure of superior SVASDs that were done from June 2018 to January 2023 at a tertiary cardiac care centre in Oman.

Case Series

The details of 7 patients are described. The following steps were in general, adopted by the authors for the successful device closure of the SVASD defects: (1) after an initial diagnosis of a SVASD by transthoracic echocardiography (TTE) and/or transoesophageal echocardiography (TEE) which were the primary screening modalities, a cardiac computed tomography (CT) angiography was performed. This was to identify additional pulmonary veins draining into the SVC above the SVASD or pulmonary veins draining below the SVASD. Cases found to have such additional pulmonary venous drainage were referred for surgical closure; (2) the procedure was performed electively. All the patients received general anaesthesia with mechanical ventilation under standard American Society of Anesthesia monitoring guidelines; (3) TEE was done prior to obtaining the vascular access; (4) the vascular access was obtained through the femoral vein, femoral artery and the right internal jugular vein; (5) it was optional to perform a diagnostic haemodynamic study for estimation of the Qp/Qs, the pulmonary artery pressure and the pulmonary vascular resistance index; (6) a superior vena cava angiography was performed for obtaining the necessary dimensions [Figure 1A]; (7) angiography of the right pulmonary artery in levophase was done to evaluate the right pulmonary veinous drainage [Figure 1B]; (8) through the arterial sheath, the left ventricle and mitral valve was crossed with a catheter to access the right upper pulmonary vein; (9) a pulmonary vein angiography was done while performing simultaneously a balloon occlusion test of the right superior vena cava [Figure 1C]; (10) a custom-made 10-Zig covered Cheatham-Platinum (CP) stent (NuMED Inc, Hopkinton, New York, USA) stent was deployed at the SVC-right atrial junction and the patency of pulmonary veins and superior vena cava was confirmed [Figure 1D]; (11) post-procedure TTE and electrocardiogram was done prior to discharge to ascertain the optimal position of the stent, and presence of pericardial effusion; and (12) antiplatelet therapy with aspirin (75 mg) for 6 months and clopidogrel bisulfate (75 mg) for 3 months was prescribed in general.

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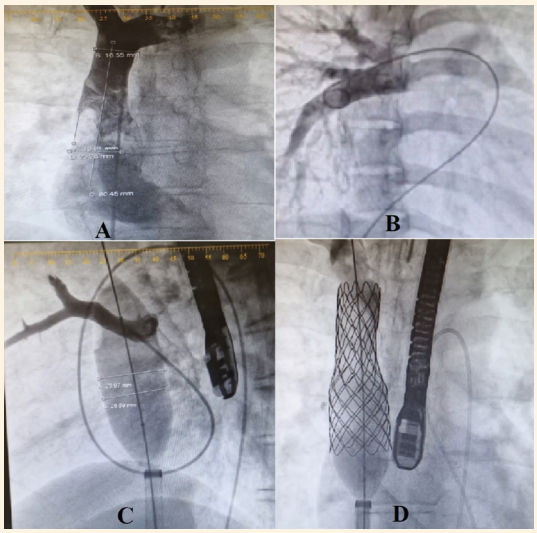

Figure 1: Image of **(A)** a superior vena cava angiography, **(B)** right pulmonary artery angiography in levophase, **(C)** balloon occlusion test with pulmonary vein angiography, **(D)** stent deployment.

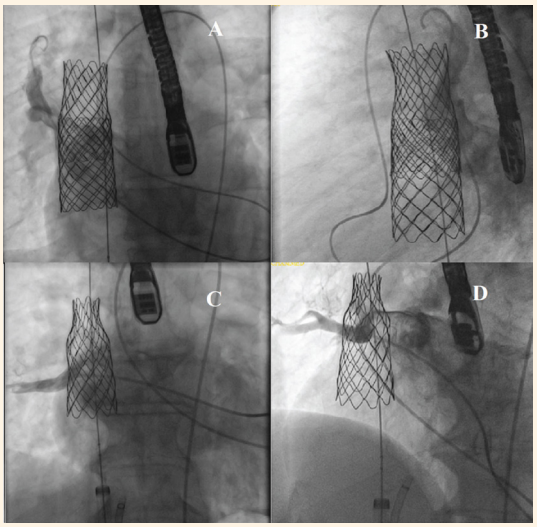

Figure 2: The anteroposterior and lateral angiograms depicting unobstructed right upper pulmonary venous drainage to the left atrium following the SVASD device deployment in **(A, B)** case 1 and **(C, D)** case 2, **(E, F)** case 3 and **(G, H)** case 4.

Institutional ethical committee approval (CR# 2023/21) was obtained for the publication of these cases.

Case 1

A 38-year-old male patient (weight = 78 kg; height = 176 cm) presented with complaints of paroxysmal nocturnal dyspnoea. The electrocardiogram showed a right bundle brach block (RBBB) pattern. TTE showed a dilated right atrium and ventricle, superior SVASD (25 mm) with a left to right shunt, anomalous right upper pulmonary vein (RUPV) draining to the superior vena cava (SVC), mild tricuspid regurgitation (right ventricular systolic pressure [RVSP]: 20 mmHg) with normal left ventricular systolic function. The Qp/Qs was 1.8. A Z-MED™ balloon (30 × 50 mm) was used for sizing the defect and two 6 cm long 10 zig covered CP stents premounted onto 26 × 60 mm and 28 × 60 mm BIB balloons were successfully deployed in January, 2023. Post-stent deployment angiography showed that the 2 stents were in appropriate position with no residual shunt and with a laminar RUPV drainage to the left atrium [Figures 2A and B; Supplementary videoclip 1]. The fluoroscopy time was 30 mins and no complications were encountered. The patient was discharged home after 2 days with the advice to take aspirin (75 mg) for 6 months and clopidogrel bisulfate (75 mg) for 3 months. TTE done at the 3-month-follow-up confirmed the correct placement of the stent with no residual shunt. The right ventricle was normal in size with a trivial tricuspid regurgitation (RVSP = 18 mmHg; tricuspid annular plane systolic excursion [TAPSE] = 28mm).

Case 2

A 47-year-old man (weight = 95 kg; height = 168 cm) presented with complaints of dyspnoea on exertion, palpitation and episodes of dizziness. TEE examination revealed a superior SVASD (18–20 mm) with a large left to right shunt and an abnormal drainage of the RUPV draining into the SVC at level of the defect. The right ventricle was dilated (basal, mid and longitudinal dimensions: 50 × 49 × 90 mm) with normal systolic function (TAPSE = 20 mm). The Qp/Qs was 1.7. A 6 cm length 10 Zig covered CP stent premounted on a BIB balloon (inner balloon 13 mm/5 cm; outer balloon 26 mm/6 cm) was deployed without any obstruction to the pulmonary venous drainage to the left atrium in January, 2022 [Figures 2C and D; Supplementary videoclip 2]. The fluoroscopy time was 38 min and there were no complications. The patient developed frequent premature ventricular contractions (PVCs) originating from the right ventricle, for which bisoprolol 5 mg was administered. He was discharged home on the 3rd day after the procedure and was advised aspirin (75 mg), clopidogrel bisulfate (75 mg) and bisoprolol (5 mg). Holter monitoring done after 2 months of the stent deployment showed low burden of PVCs and bisoprolol was discontinued. The TTE examination that was done after 8 months following the device deployment showed normal sized right and

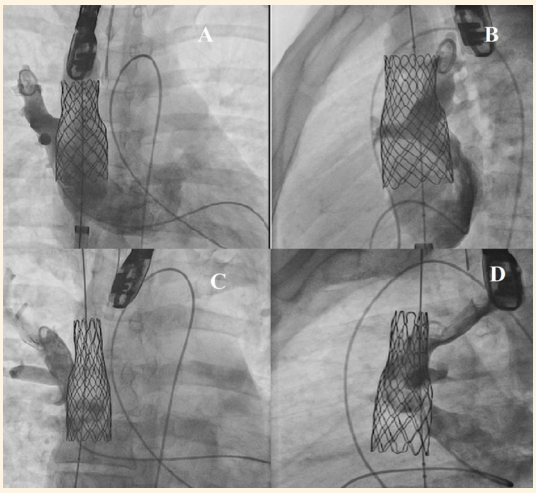

Figure 3: The anteroposterior and lateral angiograms depicting unobstructed right upper pulmonary venous drainage to the left atrium following the SVASD device deployment in (**A, B**) case 3 and (**C, D**) case 4.

left atria, with the right ventricle remaining dilated but with normal systolic function (TAPSE = 20 mm).

Case 3

A 24-year-old-female patient (weight = 71 kg; height = 152 cm) presented with atypical chest pain. TTE examination revealed a superior SVASD (25 mm) with a left-to-right shunt, moderately dilated right atrium and right ventricle (basal, mid and longitudinal dimensions: 47 × 38 × 75 mm), TAPSE was 23 mm and a mild tricuspid regurgitation. Cardiac catheterisation was done and no coronary artery disease was detected. The Qp/Qs was 1.7. A right pulmonary angiogram demonstrated a large superior SVASD (25 mm) with RUPV draining to SVC/right atrium junction. The superior SVASD was closed by a 6 cm length 10 Zig covered CP that was premounted on a 28 × 70 mm BIB balloon in June, 2018. The RUPV drainage to the left atrium was unobstructed [Figures 3A and B; Supplementary videoclip 3]. The fluoroscopy time was 32 mins with no complications. After a hospital stay of 2 days, she was discharged home with the advice to continue aspirin (75 mg) for 6 months and clopidogrel bisulfate (75 mg) for 3 months. The TTE done after 6 months of stent deployment reported the covered stent to be *in situ* with no stenosis nor turbulence in the SVC, no residual shunt, normal sized right atrium and right ventricle with normal systolic function (TAPSE = 22 mm) and a RVSP <35 mmHg.

Case 4

A 47-year-old male patient (weight = 88 kg; height = 170 cm) presented with a complaint of atypical chest pain. TTE examination revealed a mildly dilated right ventricle (basal, mid and longitudinal dimensions: 53 × 43 × 80 mm) with normal systolic function. TAPSE ~ 25 mm. The right atrium was dilated (area ~ 24 cm^2). There was mild tricuspid regurgitation, RVSP ~ 35–40 mmHg. There was a superior SVASD (30 mm). Cardiac CT confirmed a superior SVASD with the RUPV draining into the SVC. The right middle and lower pulmonary veins were draining normally to the left atrium. The Qp/Qs was 3.4. The superior SVASD was closed by a 6 cm length 10 Zig covered CP stent that was premounted on a 18 mm × 5 cm BIB without causing obstruction to the RUPV drainage into the left atrium in October, 2018 [Figures 3C and D; Supplementary videoclip 4]. The fluoroscopy time was 88 min with no complications. Patient was discharged home after 2 days with advice to take aspirin (75 mg) for life, clopidogrel (75 mg OD) for 3 months. The TTE done after 24 months following the stent deployment reported the stent to be *in situ* with no residual shunt. The right ventricle was smaller in size (basal, mid and longitudinal dimensions: 39 × 27 × 71 mm) and the RVSP was reduced (26 mmHg).

Case 5

A 47-year-old man (weight = 68 kg; height = 161 cm) presented with complaints of atypical chest pain and dyspnoea on moderate to severe exertion. TEE showed a superior SVASD (20 mm), RUPV draining into the SVC, an additional ASD secundum (5 mm), mild tricuspid regurgitation, RVSP 37 mmHg with good biventricular function. The Qp/Qs was 1.8. The superior SVASD was closed by an 8 cm length 10 Zig covered CP customized stent that was premounted on a 28 × 90 mm BIB balloon in May, 2018 [Supplementary videoclip 5]. TEE demonstrated that the RUPV drainage to the left atrium was unhindered. The fluoroscopy time was 20 min and no complications were encountered. He was discharged home after 2 days of hospital stay with advice to take aspirin (75 mg) for 3 months and clopidogrel bisulfate (75 mg) for 3 months. The TTE done after 4-years showed the stent to be in the appropriate position. The right ventricle was normal in size (basal, mid and longitudinal dimensions: 36 × 33 × 64 mm) with normal systolic function (TAPSE ~21 mm) and with a mild tricuspid regurgitation (RVSP ~24 mmHg).

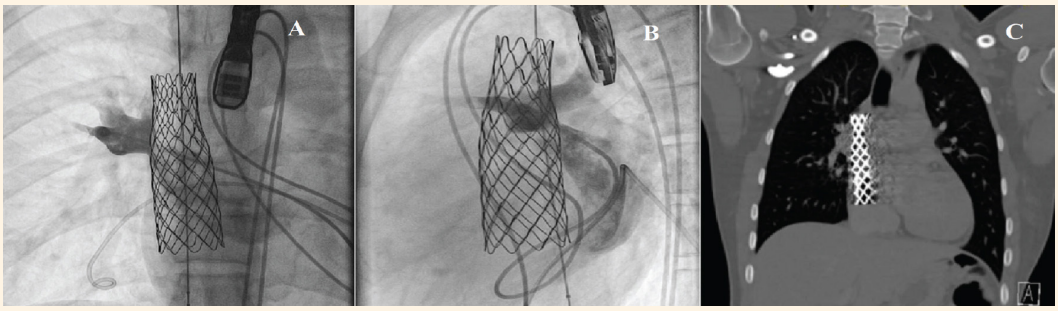

Figure 4: The anteroposterior, lateral angiograms and computed tomography angiography depicting unobstructed right upper pulmonary venous drainage to the left atrium following the SVASD device deployment in case 6.

Case 6

A 15-year-old female patient (weight = 52 kg; height = 152 cm) presented with orthopnoea. TTE examination revealed a superior SVASD (size = 16 mm), RUPV draining to the SVC, mild tricuspid regurgitation, moderately to severely dilated right ventricle (basal, mid and longitudinal dimensions: 43 × 40 × 90 mm), RVSP was 24 mmHg, dilated pulmonary artery, a normal left ventricle with an EF of 55%. The Qp/Qs was 2.0. An 8 cm long 10 Zig covered CP stent pre-mounted (on a 28 × 90 mm BIB® catheter) was deployed in October, 2021. Post deployment angiography showed the stent in appropriate position with unobstructed RUPV drainage to the left atrium [Figures 4A and B; Supplementary videoclip 6]. The fluoroscopy time was 48 mins and there were no complications. A post-procedure TEE showed no residual shunt and colour Doppler imaging showed no obstruction to the RUPV drainage into the left atrium. On the night of the procedure, the patient complained of dyspnoea. A TTE demonstrated a large mobile thrombus in the stent with a small pocket of pericardial effusion close to the right ventricular free wall. An infusion of unfractionated heparin was started and treatment with aspirin and clopidogrel was continued along with a diuretic (furosemide). TTE on the 5th day showed the resolution of the thrombus. She was discharged home on the 7th post-procedure day on rivaroxaban (15 mg) and clopidogrel bisulfate (75 mg) medication. The TTE that was done after 1-year following the deployment of the stent showed a normal sized right atrium and right ventricle, mild tricuspid regurgitation, RVSP was <35 mmHg and TAPSE was 20 mm. A CT also was done that showed unobstructed RUPV drainage with stent *in situ* [Figure 4C].

Case 7

A 56-year-old female patient (weight = 77 kg; height = 154 cm) presented with complaints of dyspnoea on exertion, giddiness and chest pain with radiation to the neck, jaw and the left arm. A TEE showed a superior SVASD (24 mm) with a left to right shunt, mild tricuspid regurgitation (RVSP = 39 mmHg), dilated coronary sinus with a persistent left SVC and an anomalous RUPV entering the lower part of the right SVC. Coronary angiography was done and coronary artery disease was excluded. The Qp/Qs was 1.7. The SVASD defect was closed with a premounted 6 cm long 10 Zig covered CP customised stent mounted on a 28 × 80 mm BIB balloon in October, 2019 [Supplementary videoclip 7]. Angiography demonstrated unobstructed RUPV drainage to the left atrium [Figures 5A and B]. The fluoroscopy time was 39 min with no complications. She was discharged home on the 2nd day with the advice to take aspirin (75 mg) and clopidogrel (75 mg). A TTE done the next day showed the stent in appropriate position with no

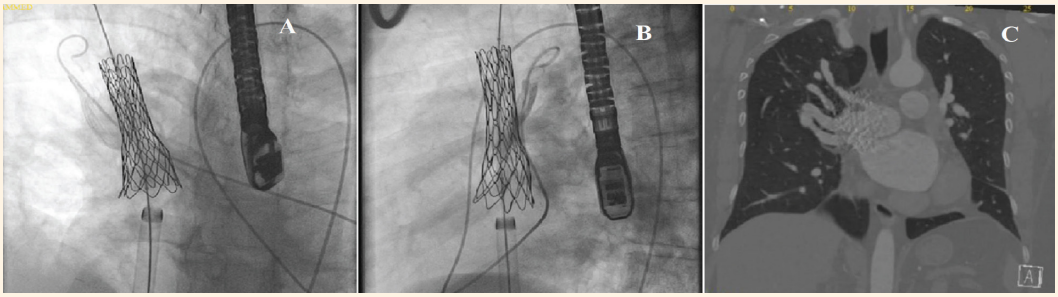

Figure 5: The anteroposterior, lateral angiograms and computed tomography angiography depicting unobstructed right upper pulmonary venous drainage to the left atrium following the SVASD device deployment in case 7.

residual shunt, normal size right ventricular (basal, mid and longitudinal dimensions: 44 × 33 × 71 mm), normal systolic function (TAPSE = 22 mm), mildly dilated right atrium (area = 18.5 cm^2), mild tricuspid regurgitation (RVSP = 35–40 mmHg). The patient was discharged home on aspirin (75 mg) for 6 months and clopidogrel bisulfate (75 mg) for 3 months. After about 12 months, a CT demonstrated unobstructed RUPV drainage to the left atrium with the stent *in situ* [Figure 5C]. An informed consent from the patients and care-givers was obtained along with institutional ethical committee approval for publication purposes.

Discussion

Transcatheter closure of SVASDs is a relatively new procedure in the field of cardiac interventions for structural heart diseases. Each institutes' experiences with the intervention may add new insights into its practicability, and the incidence and types of complications likely to be encountered. With careful planning and meticulous attention to detail, the procedure can be performed safely with a low incidence of complications. The procedure may be established as an alternative to a surgical approach in the near future.

The successful device closure of superior SVASD by interventional cardiological procedure is described. The presenting symptoms of the patients varied from various degrees of dyspnoea to atypical angina.

As SVASDs present a wide variety of anatomical features, various imaging tools, have been advocated to describe and analyse the defects. Superior SVASDs may be difficult to visualise on transthoracic echocardiography. Hence, TEE, cardiac magnetic resonance imaging and CT are frequently used for the diagnosis and for the accurate assessment of associated pulmonary venous drainage anomalies.[4] A pre-procedural multimodality imaging with 4 D flow-magnetic resonance imaging and 3D modeling were suggested for a superior outcome.[5–7] None of the patients in the current case series had a 3D modeling or 4D flow magnetic resonance done for delineating the defect. TTE and TEE were the primary investigation modalities that were used in the workup of these patients. Cardiac CT was done to exclude anomalous pulmonary vein drainage into the cephalad origin of the SVC, to plan the procedure and to rule-out coronary artery disease in elderly or symptomatic patients.

Up until recently, surgical correction was the only treatment option for SVASDs.[8] The surgical approach is associated with the risk of stenosis of SVC or pulmonary veins, residual shunting and dysfunction of sinoatrial node dysfunction.[9] Elderly patients may have a higher incidence of atrial fibrillation or flutter after surgical repair of SVASD.[1] Hence, avoidance of a surgical approach to address SVASDs when feasible appears attractive. As an alternative technique to surgery, Garg *et al*. reported a novel transcatheter approach for closure of a superior SVASD using a covered stent.[10] The technique of transcatheter correction of superior SVASDs was subsequently modified and adopted widely.[11]

Atrial flutter/fibrillation was not encountered in any patient in this current series but frequent PVCs after stent deployment was seen in one patient that responded to bisoprolol. Acute stent thrombosis with a loculated pericardial effusion occurred in another patient after the stent deployment which resolved with conservative management. In our short series, SVC stenosis, RUPV occlusion, stent embolisation and SVC rupture were not encountered in any of the cases. Incidence of stroke or vascular injuries secondary to the large sheaths used were also not encountered.

Currently available stents have certain limitations. The available devices are associated with differential shortening, a high risk of embolisation and a potential for pulmonary vein/veins erosion and obstruction. To overcome these potential shortcomings, 3 sizes were proposed to provide satisfactory closure of SVASDs with no compromise of pulmonary venous drainage.[6] The characteristics of an ideal stent are that it should be a self-expansile covered stent, distal ends should be uncovered and allow flaring to enable anchoring, middle segment should be narrower to allow unobstructed routing of pulmonary veins to left atrium, and the stents should have radiopaque markers to help appropriate positioning.[6]

In all our patients, a 10 Zig covered CP stent was deployed. This device was familiar to us as we have successfully deployed similar stents in patients with coarctation of aorta. The stent manufacturer provided a table of the foreshortening of these stents at different diameters of expansion. For device closure of SVASD defects, the stents are expanded in a filtering flask shape. The expected shortening at the site of SVC and right atrium based on their respective diameters can be predicted from this table. The stents are expanded in the SVC upto the SVC-right atrial junction; to approximately 20–30% greater than measured SVC diameter. But at the sinus venosus defect area and in the lower part of the right atrium, the stents are expanded so that it clings to the right atrial wall. For example, based on the manufacturer's table, a 6 cm stent shortens to 4.85 cm (20.20%) when it is expanded

to 26 mm diameter, to 4.39 cm (27.87%) when expanded to 28 mm diameter and to 4.11 cm (32.55%) when expanded to 30 mm diameter. The 10 Zig covered CP stent is available in custom-made lengths of 5–11 cm and is dilatable to 34 mm in diameter with moderate shortening at diameters less than 28 mm.[12] After successfully deploying the stents in the first 3 patients, we could subsequently customised the length of 10 Zig covered CP stent needed for each patient. Based on the CT angiography findings we could order a stent of customised length and diameter. However, in the first case described in this series we had to deploy two stents because the required 8 cm long stent was not available during the time of the procedure.

Deciding the length of the stent is also critical for a successful outcome in these patients. A stent that is long offers more flexibility in flaring the bottom end of the stent in order to achieve complete closure of the defect without destabilisation of the stent. Such a stent may not need an additional anchoring stent. The likelihood of stent embolisation is also minimal with long stents.[12] Careful selection of patients is critical to avoid pulmonary vein obstruction. It is also critical to interrogate the defect with a balloon catheter to mimicking the position of the stent, if it were to be deployed. When there is a suggestion of pulmonary vein obstruction during balloon interrogation of the defect, a decision needs to be made whether to go ahead and deploy the stent or abort the procedure. It is crucial to maintain a balance between over-inflation of the interrogating balloon and the stent which may lead to pulmonary vein obstruction and the under inflation which may result in a residual shunt. Placing an inflated balloon in the pulmonary vein to accord protection while the stent is inflated may be a useful precaution if a decision to proceed with the stent deployment is taken.

Deploying a stent for a superior SVASD can be challenging. Unlike in a coarctation lesion in which the stent is gripped, the cranial part of the SVC into which the stent is lodged during SVASD device closure is distensible and does not grip the stent. The prediction of the final position of the caudal part of the stent is difficult. The possibility of stent dislodgement is present as the funnel shaped stent is not always stable in the SVC. We only rely on the small waist that appears at the SVC-RA junction which is not always obvious. This becomes more challenging when the patient has bilateral SVCs as in case 4. On such occasions, the right SVC is small and flaring the lower part of the stent to obliterate any residual shunt may require multiple balloon sizes. The covered stent should not compromise the pulmonary venous drainage into the left atrium. The anomalous right upper and/or middle pulmonary veins need to be directed behind the covered stent to the left atrium. When the RUPV junction with the SVC is small, it may be a difficult to decide if a complete closure of the defect with narrowing of the pulmonary venous pathway should be opted for or to allow for an uncompromised pulmonary venous pathway with a clinically negligible residual shunt.[12]

Conclusion

We report the evolution of the transcatheter closure of superior SVASD technique at our institution. As with any new technique, modifications were continuously made. The initial experience with a relatively small number of cases has been satisfactory. The complication rate was small. The authors suggest that careful patient selection, critical planning before the procedure and meticulous assessment of the pulmonary veins during the procedure may be critical to a successful outcome.

SUPPLEMENTARY VIDEOCLIP

Videoclip 1: Angiogram showing the stent *in situ* in Case 1.

Videoclip 2: Angiogram showing the stent *in situ* in Case 2.

Videoclip 3: Angiogram showing the stent *in situ* in Case 3.

Videoclip 4: Angiogram showing the stent *in situ* in Case 4.

Videoclip 5: Angiogram showing the stent *in situ* in Case 5.

Videoclip 6: Angiogram showing the stent *in situ* in Case 6.

Videoclip 7: Angiogram showing the stent *in situ* in Case 7.

AUTHORS' CONTRIBUTION

SNM and MMM conceptualised the work. SR collected the data. SNM and MMM acquired the images. SNM, MMM, KA and AF analysed and interpreted the images. MMM drafted the manuscript. SNM, KA and AF reviewed and edited the manuscript. All authors approved the final version of the manuscript.

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