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Case Report

Unanticipated complication of transcatheter correction of superior sinus venosus atrial septal defect

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ABSTRACT

Transcatheter correction of superior sinus venosus atrial septal defect (SVASD) is being considered as an alternative to surgery in selected patients. We present the case of a 42-year-old woman with SVASD and partial anomalous venous connection of the right upper pulmonary vein (RUPV), who underwent transcatheter correction with self-expanding aortic stent graft, following feasibility assessment by balloon occlusion. Hemodynamic parameters and angiography demonstrated successful closure of the SVASD without any residual shunt and unobstructed return of RUPV to the left atrium. She developed cardiac tamponade after a few hours despite pericardial drain and underwent emergency exploratory thoracotomy. This revealed leak from a small rent in the ascending aortic wall adjacent to superior vena cava (SVC) caused by barbs of the stent protruding from SVC, without any leak in SVC. This was repaired with suture and further Teflon was placed around the barbs in SVC to prevent further injury. We also discuss the possible reason for this complication, considering our successful previous two cases with the same stents. This case highlights the importance of assessing the relationship between SVC and aorta to decide about the cranial placement of the aortic stent either by computed tomography prior or by contrast aortogram during the procedure.

Learning objective: Transcatheter correction of superior sinus venosus atrial septal defect is becoming an appropriate alternative option for cardiac surgery. There are multiple reports of this technique in the literature - mostly with balloon expandable stents, and few with self-expanding stents placed in superior vena cava (SVC). We present an unanticipated complication of self-expanding aortic stent in this situation and its management, highlighting the need for assessing the relationship between SVC and aorta to decide about the appropriate placement of stent.

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Introduction

Surgical correction has been the standard of care for patients with superior sinus venosus atrial septal defect (SVASD), until a novel transcatheter approach using a balloon expandable covered stent (BECS) deployed in the superior vena cava right atrial (SVC-RA) junction was first published in 2014 [1]. Since then various case reports and series have been published highlighting tran-

scatheter closure as a feasible and alternative option to surgery in selective patients, mostly with BECS [2–4]. However, some of these patients had to have more than one stent, either as a planned strategy or as a bail-out strategy. We believe the use of self-expanding covered stent (SECS) will avoid multiple stent usage in this situation. We present a case of transcatheter correction of SVASD with an aortic SECS and its post-procedure complication and its successful management.

Case report

A 42-year-old woman (70 kg, body mass index 26.7 kg/m²) with dyspnea for 2 months was diagnosed to have SVASD with

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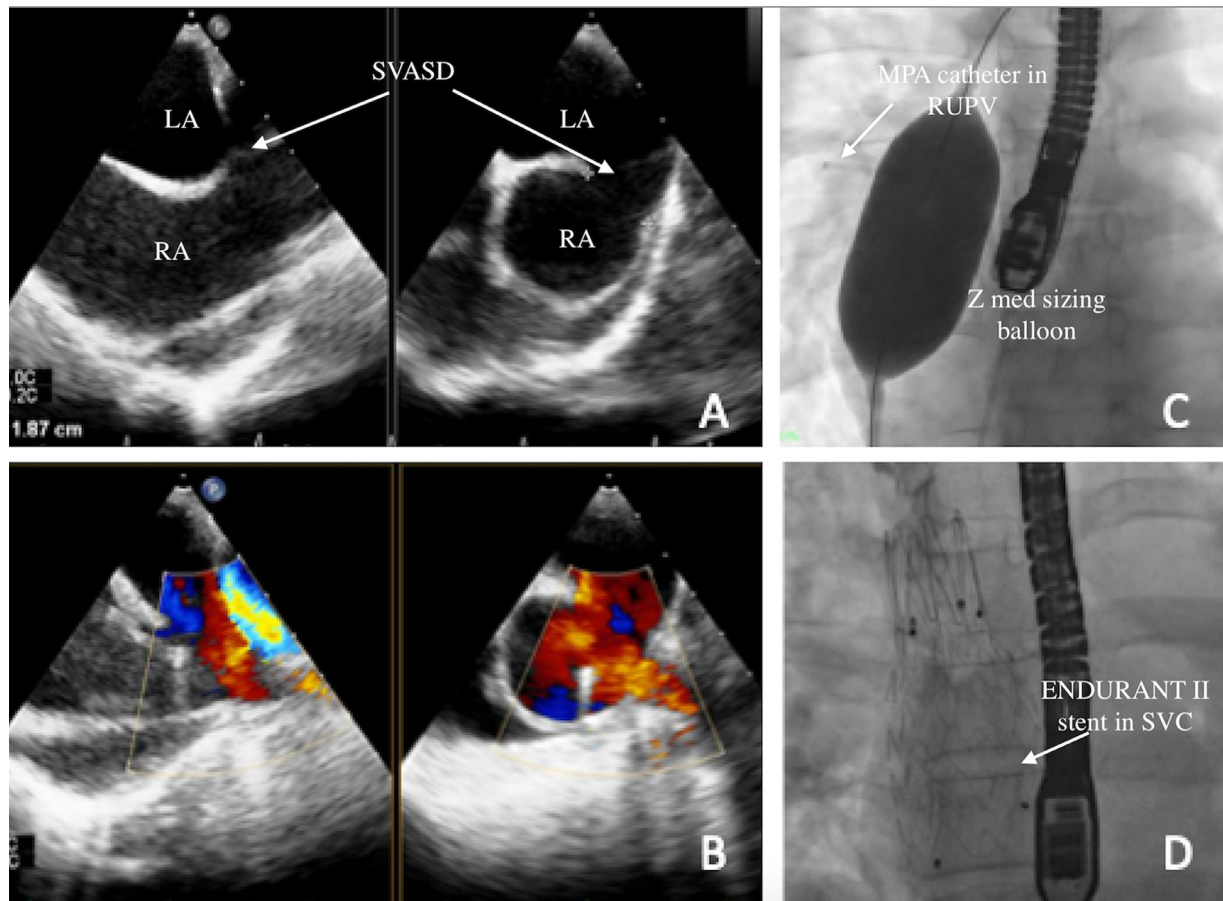


Fig. 1.

(A) TEE-biplane view, orthogonal images showing SVASD. (B) Corresponding colour Doppler image showing left to right shunt across SVASD. (C) Fluoroscopic image showing balloon occlusion with Z med sizing balloon, multipurpose catheter in RUPV. (D) Fluoroscopic image showing Endurant II stent graft post deployment in SVC.

LA, left atrium; PFO, patent foramen ovale; RA, right atrium; RV, right ventricle; RUPV, right upper pulmonary vein; SVASD, sinus venosus atrial septal defect; SVC, superior vena cava; TEE, transesophageal echocardiogram.

dilated right-sided chambers, large left to right shunt ($Q_p:Q_s$ ratio 3:1) and normal biventricular systolic function on transthoracic echocardiogram (TTE). Transesophageal echocardiography (TEE) showed defect size of 25.5 mm (Fig. 1A,B, Video 1) with right upper pulmonary vein (RUPV) draining into the right atrium. Under fluoroscopic and TEE guidance, assessment for feasibility of transcatheter correction was performed with balloon occlusion of SVC using Z Med 30 × 50 mm balloon (Braun Med Inc., Melsungen, Germany) on an Amplatzer super stiff wire (Boston Scientific Corporation, Natick, MA, USA) via 18F sheath in right femoral vein (Fig. 1C). The balloon was inflated in a way that it completely covered the upper end of RUPV-SVC junction and the lower end of the balloon completely occluded the defect. The selective RUPV hand injection was done using 5F Judkins catheter, which was placed earlier from the left femoral vein, via patent foramen ovale (PFO) and left atrium (LA) into RUPV. Unobstructed RUPV flow into LA without stasis of contrast and no rise in pressure was also confirmed using fluoroscopy and TEE imaging simultaneously. Subsequently, she underwent transcatheter correction of SVASD using 32 × 49 mm ENDURANT II aortic stent graft (Medtronic, Minneapolis, MN, USA) in SVC accessed via 18F sheath in right femoral vein (RFV) under general anesthesia (Fig. 1D). Post stent deployment RUPV angiogram was done to show unobstructed flow in RUPV and no residual shunt (Video 2). TEE also confirmed no residual shunt with RUPV flow into LA (Fig. 2A and

B, Video 3). Stent position was also confirmed with both TEE and computed tomography (CT) scan (Fig. 2C and D, Video 4). She was extubated immediately post procedure and was hemodynamically stable. Surveillance TTE done prior to transfer from the cardiac care unit (CCU) after 4 hours showed stent in-situ, with no residual shunt and pericardial effusion. Nine hours post-procedure, she suddenly developed back pain and was found to be tachycardic (106 bpm) with blood pressure of 80/60 mmHg. Repeat TTE showed large pericardial effusion with signs of cardiac tamponade. She underwent emergency pericardiocentesis and 200 ml of hemorrhagic fluid was aspirated. Her clinical and hemodynamic parameters improved. Pig-tail catheter was left in-situ and monitored in CCU, with minimal inotropic support. Her hemodynamics worsened again after 3 hours and a repeat TTE showed recurrent pericardial collection despite continuous drainage. The cardiac surgical team was involved for emergency exploratory thoracotomy, which revealed SECS anchoring pins or barbs (Fig. 2E) protruding out through SVC, scraping the adjacent ascending aortic wall with a tiny rent in the ascending aorta. There was no leak from the SVC. The rent in the aorta was sutured and Teflon felt was placed around the SVC at the level of the barbs to avoid further injury to the aorta (Fig. 3). Repeat TTE after a few hours showed stent in-situ with no pericardial effusion. She was discharged with dual antiplatelet therapy after 4 days in a stable condition.

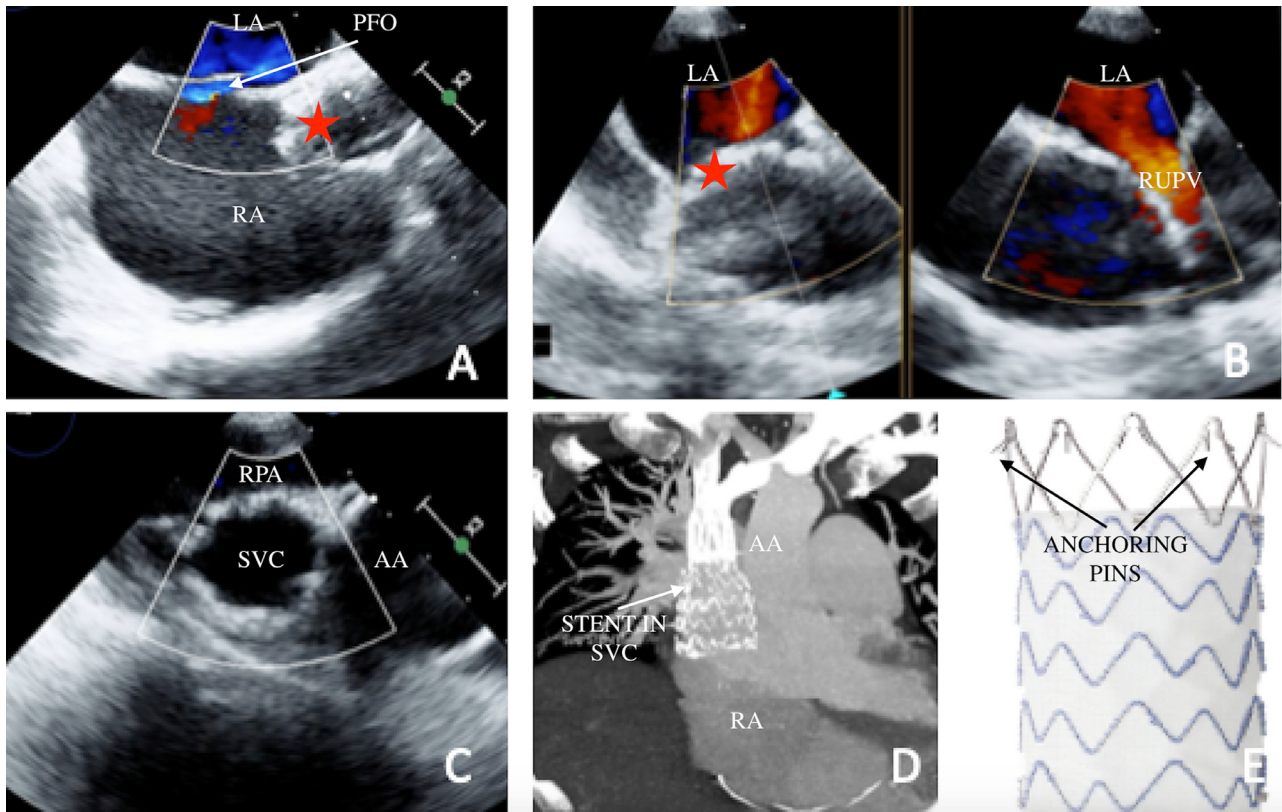


Fig. 2.

(A) TEE- Bicaval view showing stent (red star) from SVC to RA. Colour Doppler shows small left to right shunt across PFO with no residual shunt across SVASD. (B) Biplane view, orthogonal images showing RUPV flow directed into LA (red star showing stent in SVC). (C) TEE showing the stent in SVC and its proximity to aorta and RPA. (D) Cardiac computed tomography in the coronal view showing stent in SVC and its proximity to aorta. (E) ENdurant II aortic graft with anchoring pins. AA, ascending aorta; TEE, transesophageal echocardiogram; LA, left atrium; PFO, patent foramen ovale; RA, right atrium; RUPV, right upper pulmonary vein; SVASD, sinus venosus atrial septal defect; SVC, superior vena cava; RPA, right pulmonary artery.

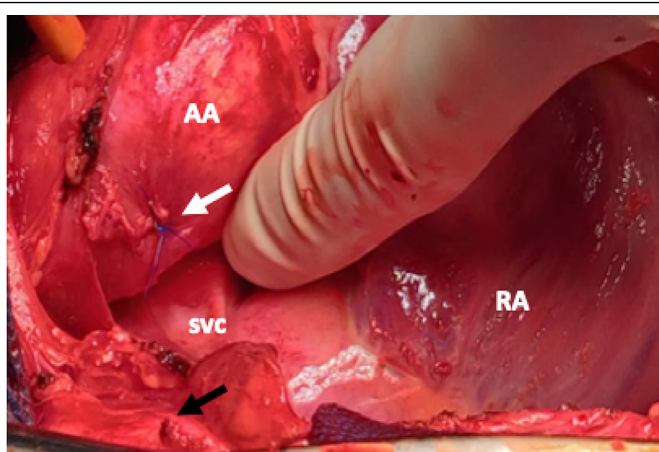


Fig. 3.

Intraoperative image, surgeon's view showing suture on AA. Black arrow head showing barbs protruding out of SVC. AA, ascending aorta; RA, right atrium; SVC, superior vena cava.

Discussion

Further to the first case report of successful transcatheter closure of SVASD [1], various technical iterations have emerged both for assessment and treatment. Although most operators have used BECS in this situation previously [2–4], there have been various

concerns such as having control over the stent expansion (size mismatch at the SVC-RA level) leading to mal-apposition, stent migration, and need for multiple stents [2]. Even in case series with prior 3-D assessment and planning, 52% (13 out of 25 patients) had additional stents used and 36% (9 out of 25) of the patients had to have a second stent for anchoring the BECS. During flaring up of the RA end of the stent, migration into RA was noted in 5 of these 9 patients and an additional uncovered anchoring stent had to be used to stabilize the SVC end [2]. In the latest series with BECS, 9 out of 24 (38%) patients had more than one BECS. Stent migration was noted in three patients, out of which two had to undergo cardiac surgery [4]. The first report of SECS usage in this condition was in a 23-year-old female with zenith flex AAA Endovascular stent graft (Cook Medical, Bloomington, IN, USA), where three stent grafts were used as a planned procedure [5]. The successful use of single SECS Endurant II aortic graft was reported in 2018 in a 63-year-old man, where the authors raised concerns about the barbs in the SVC leading to perforation (unlike the thick-walled aorta where the stent graft is being used routinely for interventions) [6]. The advantage of SECS is it adapts to the structure it is deployed in and does not need additional flaring at the RA end to cover the defect as is the case with BECS. The anchoring pins have an added advantage of preventing stent migration, thereby avoiding additional stents as noted in previous reports. Also, obstruction to the pulmonary venous flow during balloon dilation of RA end and prophylactic placement of inflated balloon in the RUPV was noted in the previous series, which we have not experienced with the three cases of SECS until now [2].

We have used the same Endurant II aortic graft SECS successfully, 6 and 4 months earlier in a 27-year-old and a 44-year-old male respectively with single stent graft. They had 70-mm long SECS and are doing well without any complications. Their follow up CT did not show any evidence of RUPV obstruction with normal drainage into LA. This is our third patient, where a 49-mm long SECS was used. As described earlier in this report, there was no leak noted from the SVC despite the barbs protruding out during exploratory thoracotomy, possibly due to the low pressure in SVC. The proximity of SECS barbs to the aorta in this patient is different from the other two patients. Since the distal part of the stent is at the RA level in all three patients, the location of the cranial part of the stent in this patient (49 mm) is at lower level compared to the other two patients (70 mm). We decided to choose a 49-mm long stent in this patient after fluoroscopic evaluation, as we believed a shorter stent will serve the purpose, as it did when evaluated post deployment. However, retrospectively we realized that the barbs in the previous two patients were higher up in the SVC where the ascending aorta courses away from the SVC, while in this patient, the barbs were too close to the aorta. We believe the injury to the aorta in this patient is due to this close proximity and happened only when the patient started to mobilize with changes in hemodynamics. This is the possible explanation of her deterioration a few hours after the procedure. Another probable reason can be the slow expansion of the graft over a few hours after deployment leading to barbs projecting out of the SVC and perforating the aorta.

We present this case to highlight the complication of using SECS with barbs in the SVC which is routinely used for aortic interventions. This case also highlights the importance of assessing the relationship between SVC and aorta to decide about the cranial placement of the SECS either by CT prior to the procedure or aortogram during the procedure. By addressing these issues, sin-

gle SECS with aortic stent graft with barbs can be considered as a viable alternative for this situation rather than multiple BECS.

Declaration of Competing Interest

All authors declare that there is no conflict of interest.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.jccase.2021.07.005](https://doi.org/10.1016/j.jccase.2021.07.005).

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