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Long-Term Follow-Up after Percutaneous Closure of Large Aortic Root Pseudoaneurysm with a Flex II Atrial Septal Defect Occluder: A Case Report

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Abstract

Pseudoaneurysm of the aortic root is a rare but potentially lethal complication after surgical manipulation of the aorta, trauma, or infection. Open repair is currently the standard of care, but it is associated with high morbidity and mortality. Recently, percutaneous treatment has emerged as an alternative to surgery in selected cases. Here, we report our experience of successful percutaneous closure of a large aortic root pseudoaneurysm in a male patient with a previous mechanical aortic valve replacement and concomitant aortoplasty.

Keywords: Pseudoaneurysm of the aortic root; Percutaneous closure; Atrial septal defect occluder; Transesophageal echocardiography; Cardiac computed tomography; Case report

Introduction

Aortic Root Pseudoaneurysm (ARPA) is a life-threatening complication following cardio-thoracic surgery. In cases where surgical correction is deemed too high-risk [1,2] and stent coverage not feasible, percutaneous exclusion of aortic pseudoaneurysm [3] can be considered using embolization of coils [4] and local thrombin injection [5]. Another endovascular option is the off-lable use of septal occluder devices [6-9]. We report the long-term follow-up result after catheter-based closure of a large ARPA using a Flex II Atrial Septal Defect Occluder (F II ASDO, Occlutech, Helsingborg, Sweden).

Case Presentation

This is the case of a 57-year-old man with a history of salicylate allergy, arterial hypertension, stage III chronic kidney disease, pulmonary silicosis, autoimmune reactive arthritis, depression and ascending aorta (AA) aneurysm with severe aortic regurgitation on furosemide 50 mg daily, ramipril 2.5 mg daily, bisoprolol 5 mg daily, simvastatin 20 mg daily, potassium canrenoate 50 mg daily and prednisone 6.25 mg daily.

He was surgically treated in 2011 with a mechanical mono-disk aortic valve implantation (St Jude Medical 25 mm) and concomitant aortoplasty. In May 2017 the patient was admitted for dyspnoea, cough and hyperpyrexia with a clinical suspect of acute pneumonia. ECG showed sinus tachycardia. Chest X-ray confirmed the presence of bilateral basal pulmonary infiltrates, diffuse interstitial thickening and widening of mediastinic silhouette. Blood test showed increased white blood cell count (12.5 × 109/L), high CRP (7 mg/dL) and creatine (2.2 mg/dL), mild anemia (Hb 11.8 g/dl). Blood cultures and serological tests were negative for chlamydia and mycoplasma. Contrast enhanced Computed Tomography (CT) was performed confirming the diagnosis of multifocal interstitial pneumonia. Moreover, the contrast injection incidentally showed a giant ARPA, originating from non coronary sinus of Valsalva and characterized by concentric thrombotic stratifications (maximum outer diameter of 61 mm; luminal diameter of 24 x 40 mm; collar of 6 x 7 mm; distance from the aortic valvular plane of 6.5 mm) (Figure 1). Two-dimensional (2D) transthoracic (TTE) (Figure 2) and three-dimensional (3D) transesophageal echocardiography (TEE) (Figure 3) confirmed the presence of ARPA and showed normal aortic prosthesis performance, a preserved left ventricular ejection fraction, and lastly a dilatation of aortic root (50 mm) and AA (47 mm). No significant coronary artery stenosis was angiographically shown. After heart team discussion and in consideration of the high surgical risk (logistic Euro-score: 29.54%; Euro-SCORE II: 7.65%), it was decided to address his ARPA percutaneously using an atrial septal defect device in an off-label way so as to seal the ARPA orifice. Written informed consent, after explanation, was obtained from the patient. The patient received for 20 days broad spectrum antibiotic therapy with meropenem, caspofungin and vancomycin until the date of procedure.

The procedure was performed under general anesthesia and 2D/3D TEE guidance. Right and left femoral accesses were obtained with 8F and 6F short introducer sheaths, respectively. A 6-Fr pigtail catheter was advanced from the left femoral artery in AA for a baseline angiography (Figure 4) in two orthogonal projections and it was left in place to guide the procedural steps of the procedure. A 6-Fr internal mammary artery (MA) guiding catheter with hydrophilic-coated 0.035" guide-wire was used to engage the ARPA collar and go inside the lumen of the pseudoaneurysm. The hydrophilic wire was replaced by a super stiff 0.035" 260 cm "J" tip guide wire, preformed with small curve, that was pushed into the lumen of ARPA (Supplementary Video S1). Due to the severe tortuosity of the ileofemoral axis and the unfavourably sharp angulation of ARPA entry orifice in relation with the AA wall, we failed to reach the ARPA cavity using a 7-Fr, 45°curved, 110 cm long sheath. We then changed for a telescopic system, made of 8-Fr straight 90 cm long sheath and 6-Fr MA guiding catheter, but we lost again the access to the ARPA collar. In order to improve the backup support, we shifted to a new telescopic system, formed by 12-Fr right groin introducer 30 cm long and 8-Fr, 45°curved, 110 cm long sheath (Figure 5A and B). This system allowed a deep engagement of the ARPA. Finally, a 7.5 mm F II ASDO with a distal disc of 18 mm and a proximal disc of 14 mm was selected. After opening the two discs across the ARPA collar (Figure 5C), we performed a stability test and finally the device was released (Figure 5D).

Post-procedure 2D/3D TEE color Doppler and AA angiography (Figure 6, Supplementary Video S2) documented a complete closure of ARPA without interference with mechanical valve or coronary artery. The patient's hospitalization course was eventless except for multifactorial anemia requiring blood transfusion. Follow-up 2D TTE color Doppler at 48 hours and 15 days after the procedure showed a correct and stable position of the device with complete closure of the ARPA. No vascular complications or device migration occurred within the follow-up period. Three years later, he remained symptom free and TTE showed the correct position of the device. Notwithstanding, the patient died four years after the procedure in multiorgan failure due to a no-device related septic shock.

Discussion

ARPA is usually a late complication of previous surgical aortic valve replacement and coronary artery bypass graft. ARPA may occur at the site of cannulation or aortotomy and is associated with unpredictable rate of expansion, risk of rupture or fistula formation, thrombosis with distal embolization and death, if left untreated. For patients deemed at high risk or in whom conventional surgery is not possible, endovascular procedures can offer a less invasive and life-saving alternative.

There are different percutaneous options for closure of aortic pseudoaneurysms such as the use of stent grafts [10], coil embolizations [11], thrombin injection in the sac [5]. Use of off-label septal occluder devices, mostly Amplatzer ASD device, has been described in various case reports (6-9) and has shown better results.

In our case, we used for the first time a F II ASDO, a self-expanding, single hub double-disc device made from nitinol-braided wires [12]. Product competitive features are an optimized braiding pattern and less material at left atrial disc (no hub). Moreover, Flex II delivery system has an improved design that provide better flexibility and a smaller delivery sheath. The unique ball-connection between the Flex II pusher and the occluder safely locks it, while it freely follows the anatomy with a 50° angle (Supplementary Figure S1) allowing superior adaptability, better alignment and a more physiological implant position of the device as demonstrated in our case. Futhermore, during deployment the left atrial disc develops in a round, ball-like shape omitting the flat profile of other devices with a double-sided hub: this prevents a prolapse of the left-sided disc during the implantation process.

Nevertheless, the experience of catheter-based closure of pseudoaneurysm of the ascending aorta gained so far is still limited and individualized approach to treatment is mandatory. Technical challenges need to be considered in the performance of these complex procedures, including adequate device selection and appropriate sizing /curves of guiding catheters for device delivery in order to avoid further serious morbidities such as device embolization.

Finally, appropriate pre-procedural planning and multimodality imaging such as 2D/3D TEE and CT are mandatory for anatomic assessment and might be key for success.

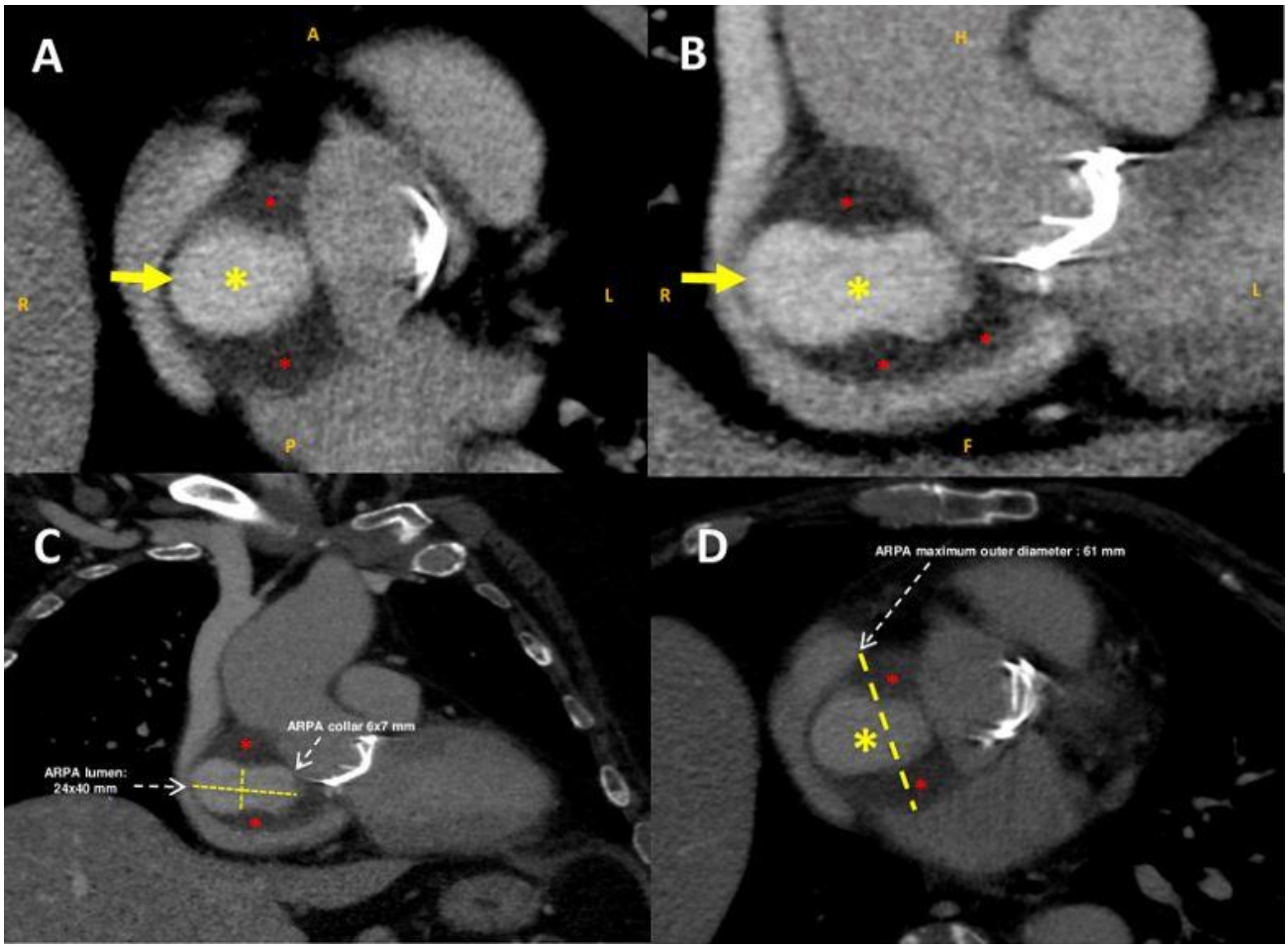


Figure 1: Contrast enhanced CT of the thoracic aorta. ECG gated axial **(A)** and coronal **(B)** volume-rendered images show a large Aortic Root Pseudoaneurysm (ARPA) (yellow arrow) arising from the previous aortic suture whit a large inner cavity (yellow star) and concentric thrombotic stratifications (red stars). ECG gated axial **(C)** and coronal **(D)** volume-rendered images summarize the main morphological characteristics of the ARPA and the relationship with the adjacent structures.

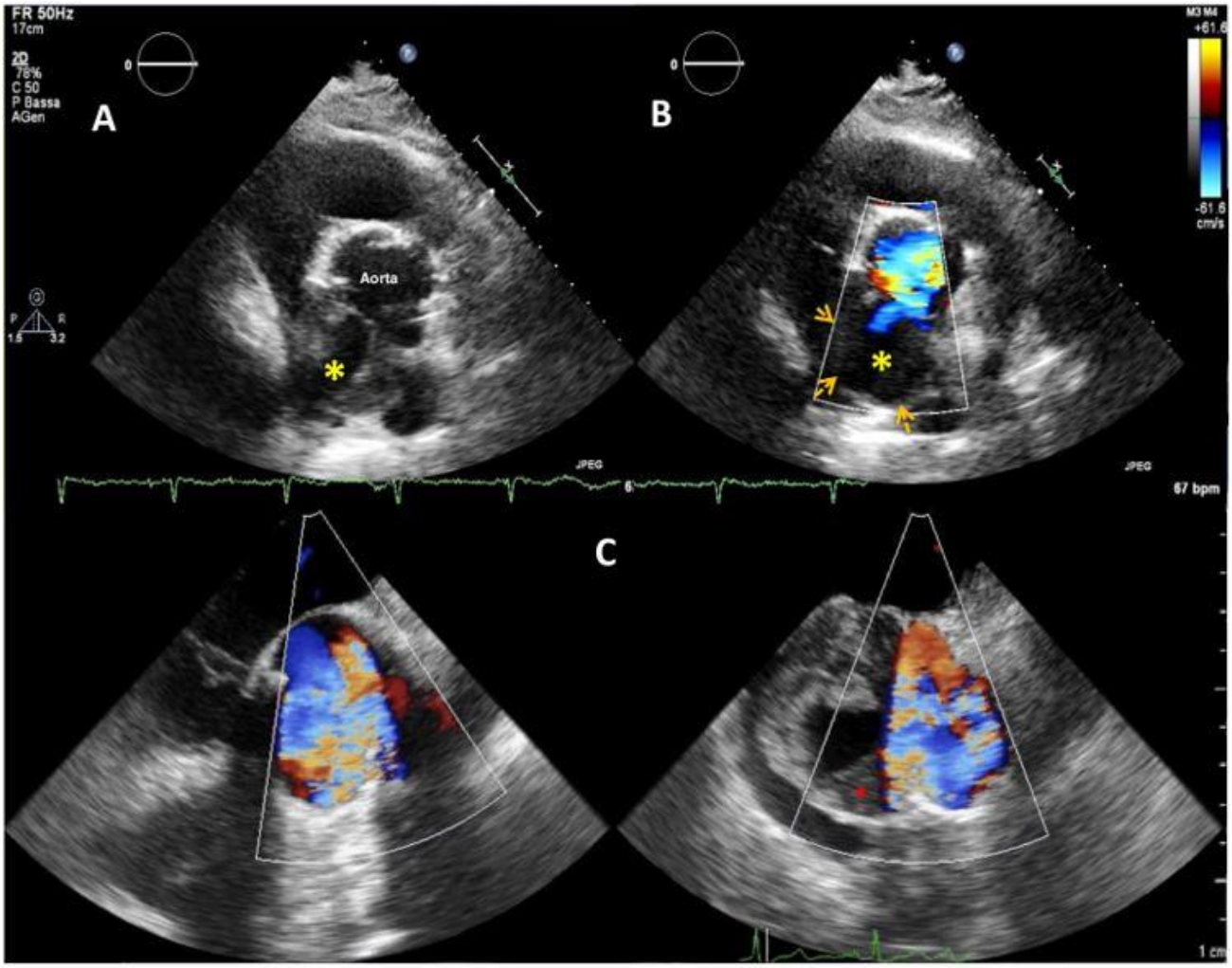


Figure 2: 2D TEE middle esophageal short axis view of aortic root showing the ostium of a giant ARPA (yellow star), originating from non coronary sinus of Valsalva **(A)** and color flow inside ARPA (borders outlined by orange small arrows) in the systolic frame **(B);** 3D TEE color Doppler X-plane rendering middle esophageal view showing the giant ARPA with thrombotic apposition (red star) and color flow inside aortic root and ARPA ostium.

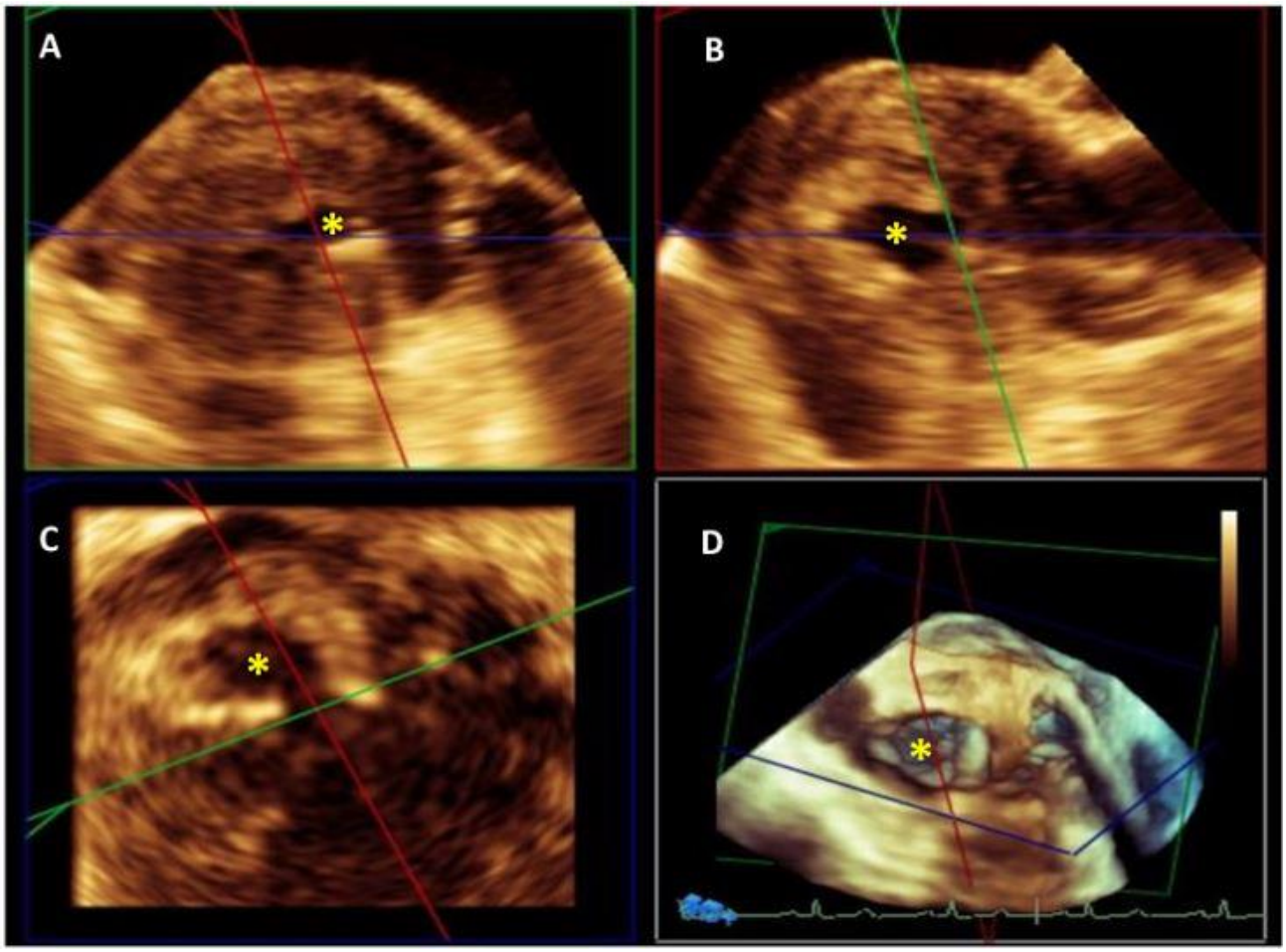


Figure 3: 3D TEE middle esophageal view Multiplanar Reconstruction rendering of the ARPA (yellow star) in the transversal **(A),** longitudinal **(B)** and coronal planes **(C);** 3D TEE ARPA volume rendering **(D).**

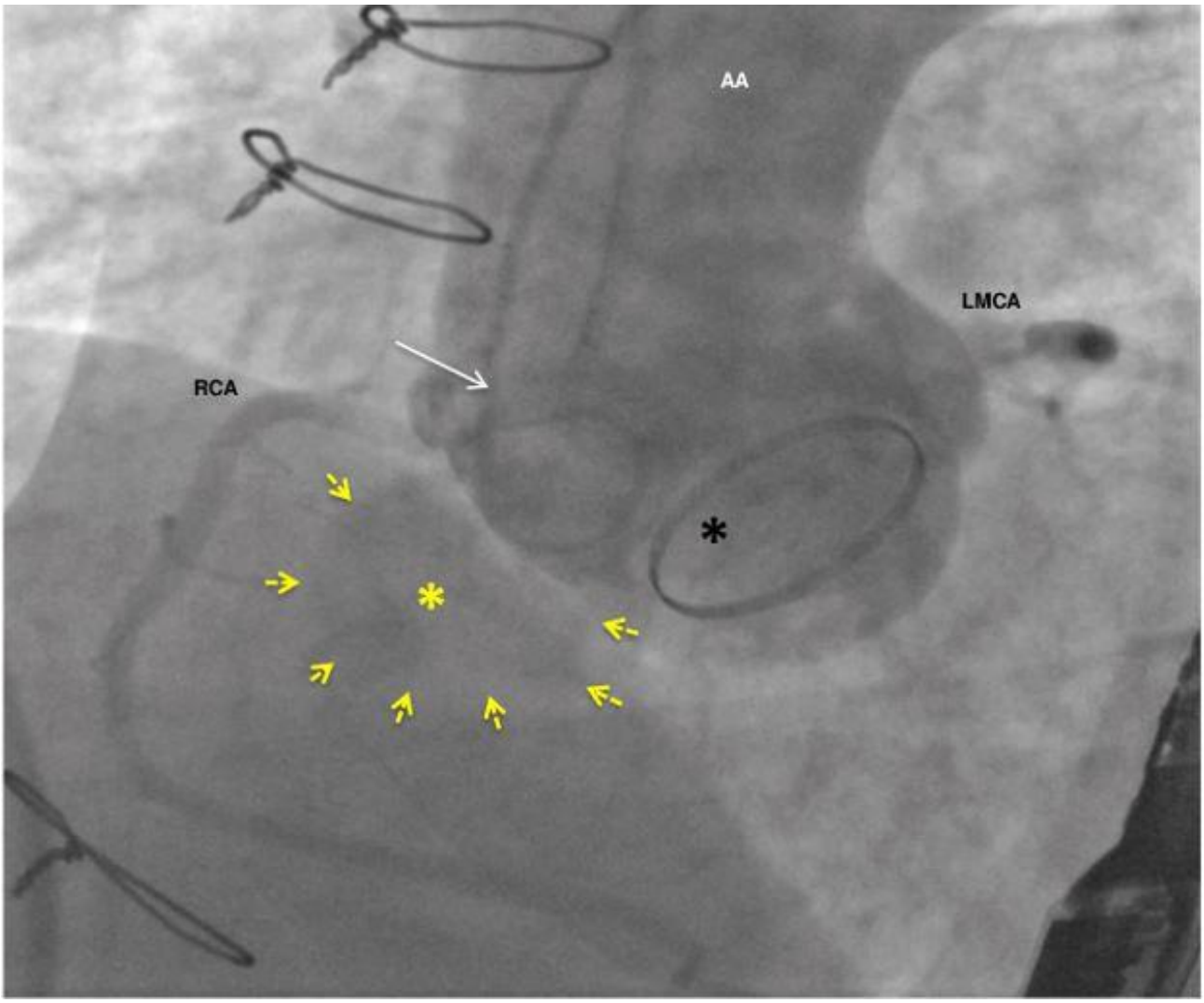


Figure 4: Baseline ascending aorta angiography in Left Anterior Oblique (LAO) projection showing the ARPA (yellow star) adjacent to the bioprosthetic aortic valve (black star). The borders of ARPA are delineated by the yellow small arrows. The white arrow indicates the pig-tail angiographic catheter.

LMCA: Left Main Coronary Artery; RCA: Right Coronary Artery; AA: Ascending Aorta

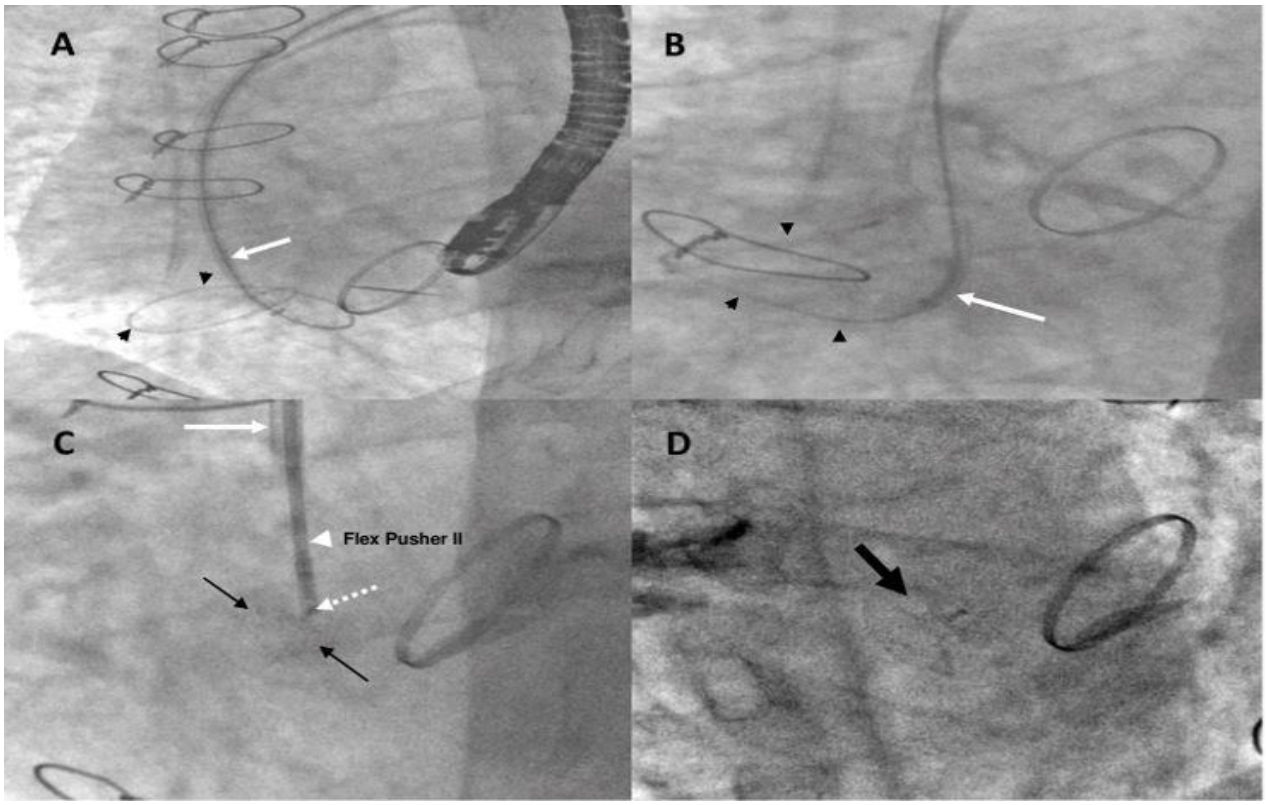


Figure 5: Fluoro-angiographic procedural steps. **A:** a super stiff 0.035" 260 cm "J" tip guide wire (black arrowhead), preformed with small curve, pushed into the lumen of ARPA while the 8F, 45° curved, 110 cm long sheath (white arrow) is not yet engaged in the ARPA entry orifice; **B:** the long sheath (white arrow) better aligned and engaged into the ARPA collar; **C:** the Flex II pusher (white arrowhead), a bioptome-like delivery system, is still attached to the ball (hub) (white dashed arrow) of the proximal disc of the Flex II ASD occluder (black arrows); **D:** the 7.5 mm Flex II ASD occluder (black arrow) finally deployed in the correct position across the ARPA collar.

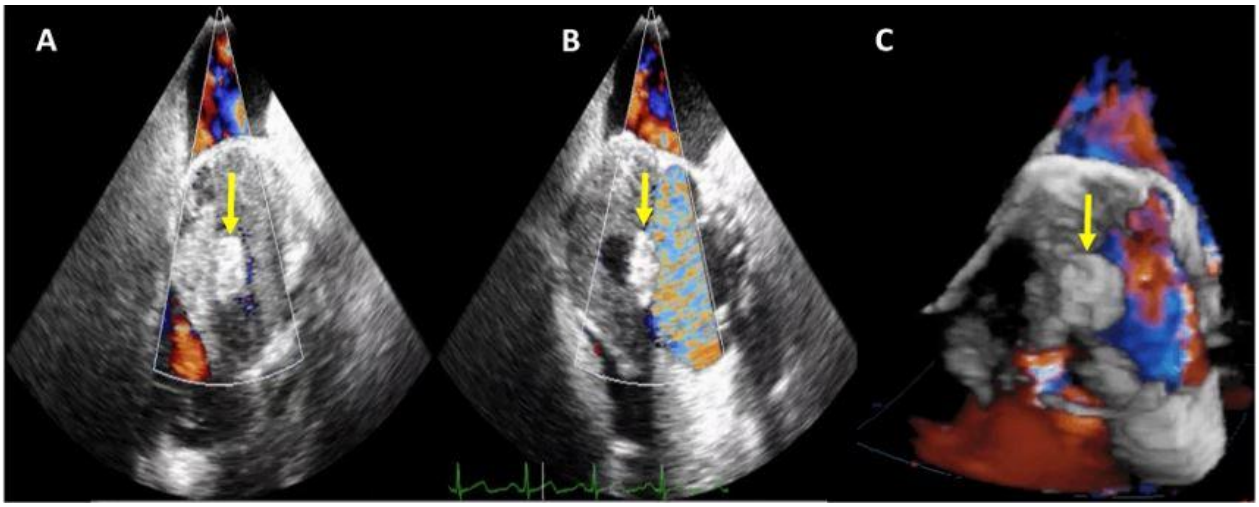
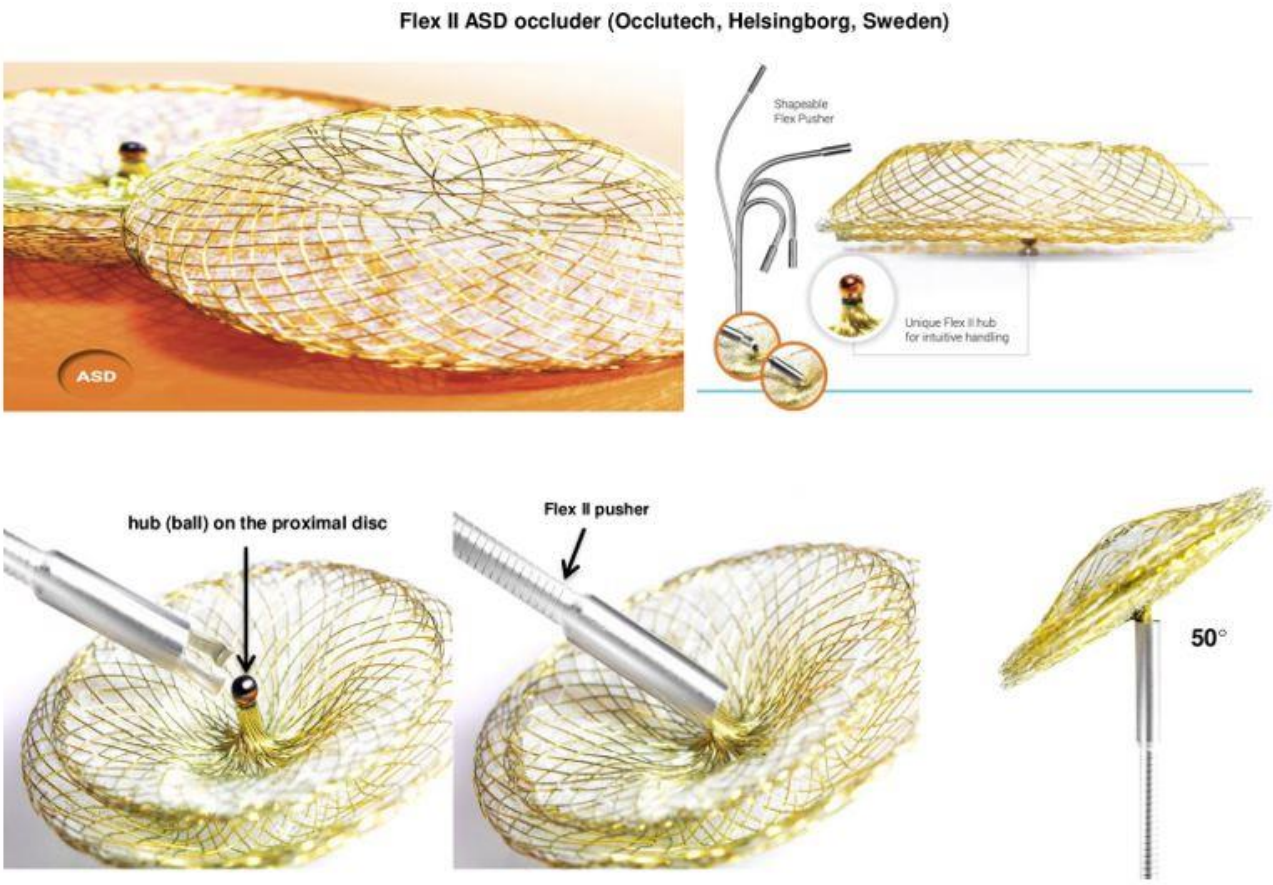


Figure 6: Post-procedure 2D TEE middle esophageal color Doppler view **(A and B)** and 3D TEE volume crop rendering **(C)** showing the device (yellow arrow) perfectly positioned with no evidence of flow inside the ARPA.

Supplementary Materials



Supplementary Figure S1: The Flex II ASD occluder (Occlutech, Helsingborg, Sweden) is made out of a set of nitinol wire braids secured by a single hub, allowing more pliability with a superior adaptability. A hubless distal disc minimizes the amount of material. The single hub (ball) is on the proximal disc. The unique ball-connection between the Flex II pusher and the occluder safely locks it, while it freely follows the anatomy with a 50° angle allowing superior adaptability, better alignment and a more physiological implant position of the device.

Supplementary Video S1: 2D X-plane Transesophageal Echocardiography (TEE) showing a super stiff 0.035" 260 cm "J" tip guide wire, preformed with small curve, pushed into the lumen of ARPA.

Supplementary Video S2: 2D TEE color Doppler showing the Flex II ASD occluder correctly placed across the collar of ARPA without residual shunt.

Note: *Supplementary videos are related to this article can be found online at:

https://www.literaturepublishers.org/archive/Long-Term-Follow-Up-after-Percutaneous-Closure-of-Large-Aortic-Root-Pseudoaneurysm-with-a-Flex-II-Atrial-Septal-Defect-Occluder:-A-Case-Report.html

Conclusion

The use of Flex II ASD occluder as an off-label way so as to seal the ARPA orifice represents an effective less invasive alternative to surgical repair with the aim of protecting the patient from life-threatening complications.

Based on the properties described, we believe that this occluder system is a step forward in the interventional armamentarium for aortic pseudoaneurysms closure.

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Author's Contributions: Vercellino M, Costante AM, and Onorato EM drafted the initial manuscript. Onorato EM, Vercellino M, Reale M and Bartorelli AL critically reviewed the manuscript. All authors approved the final manuscript.

Teaching Points

1. To understand the role of multimodality imaging approach for location and sizing of the aortic root pseudoaneurysm.
2. To review the equipment options and techniques, including use of catheters, sheaths, closure devices for this uncommon but life-threatening complication following cardio-thoracic surgery.

Conflict of Interest: Eustaquio Maria Onorato is a consultant for Occlutech, manufacturer of the device. The remaining Authors declare no commercial or financial relationships that could be construed as a potential conflict of interest.

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