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CASE REPORT

Cusp thrombosis of a self-expandable sutureless aortic valve treated by valve-in-valve transcatheter aortic valve implantation procedure: case report

Ulvi Cenk Oezpeker[1]*, Gudrun Feuchtner[2], and Nikolaos Bonaros[1]

[1]University Clinic of Cardiac Surgery, Medical University of Innsbruck, Anichstrasse 35, 6020 Innsbruck, Austria; and [2]University Clinic of Radiology, Medical University of Innsbruck, Anichstrasse 35, 6020 Innsbruck, Austria

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Background Surgical implantation rates of bioprosthetic valves, especially the use of sutureless or rapid deployment valves, as well as the advent of transcatheter valve implantation (TAVR) have increased during the last decades mainly due to their excellent haemodynamic and clinical results. One common characteristic of all bioprosthetic types of surgical aortic valve replacement (SAVR) and TAVR is the risk of early degeneration, which leads to valve-dysfunction and is associated with higher rates of valve reinterventions. Recent studies have demonstrated that cusp thrombosis may play a role in early valve dysfunction. This case report is, to our knowledge, the first documentation on a successful treatment of early aortic valve (AV) degeneration of a sutureless AV thrombosis with a valve-in-valve (ViV) TAVR implantation.

Case summary A 77 years old woman was re-evaluated from the heart-team, which considered the following characteristics: severe impairment of mobility and frailty with an STS-score of 10.01% and a EuroSCORE II of 6.9%. Due to the high surgical risk for SAVR, we decided to perform a ViV-TAVR using a balloonexpandable bioprosthesis. The procedure was performed via transfemoral access under general anaesthesia using a 23 mm Edwards-Sapien 3 bioprosthesis without balloon-valvuloplasty and with nominal-volume dilation under rapid-pacing.

Discussion The differentiation of bioprosthesis valve thrombosis, and hypoattenuating leaflet thickening vs. structural valve degeneration can be difficult, and a multimodality imaging approach, comprising trans-thoracic echocardiogram, trans-oesophageal echocardiography and computed tomography, useful. These investigations are very important to decide the right strategy of surgical valve replacement vs. TAVR.

Keywords Case report • Sutureless valve • Bioprosthesis thrombosis • Valve in valve TAVI

Learning points

- Cusp thrombosis of a bioprosthesis may also affect sutureless valves and is probably related to an elliptic expansion of the prosthesis.
- Transcatheter valve-in-valve implantation can be used as a therapeutic option in case of failure of anticoagulation treatment.
- A combination of haemodynamic measurements and multimodality imaging is essential for the diagnosis of bioprosthetic cusp thrombosis and the evaluation of treatment success.

* Corresponding author. Tel: +43 50504 83415, Fax: +43 50504 22502, Email: cenk.oezpeker@tirol-kliniken.at
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Introduction

Surgical implantation rates for bioprosthetic valves, especially sutureless or rapid deployment valves, as well as the advent of transcatheter valve implantation (TAVI)[1] have increased during the last decades mainly due to their excellent haemodynamic and clinical results.[2] One common characteristic of all bioprosthetic types of surgical aortic valve replacement (SAVR) and TAVI is the risk of early degeneration,[3] which leads to valve dysfunction and is associated with higher reintervention rates. Recent studies have demonstrated that bioprosthetic valve thrombosis (BVT) may play a role in early valve dysfunction. This case report is, to our knowledge, the first documented report on therapy of a BVT of a sutureless valve (SV) with a valve-in-valve (ViV) TAVI.

Timeline

Symptom onset	November 2015
Diagnosis of aortic stenosis	January 2016 to February 2016
Surgical aortic valve replacement	March 2016
Recurrence of heart failure symptoms	February 2017
Cusp thrombosis	March 2017
Failure of pharmacological thrombus resolution	May 2017
Valve-in-valve transcatheter valve implantation	June 2017
Last follow-up	March 2018

Patient information

A 77-year-old woman presented at our department for treatment of a highly symptomatic low-flow (SVI: 23 mL/m^2/1.73 m^2 body surface area), low-gradient (mean: 38 mmHg) AV stenosis (AV area 0.52 cm^2) with preserved ejection fraction (left-ventricular ejection fraction (LVEF): 58%), and a very hypertrophic left ventricular myocardium. In addition, she suffered from secondary pulmonary hypertension (catheter-measured systolic pulmonary artery pressure 50 mmHg). At that time she was dyspnoeic at New York Heart Association (NYHA) functional status III. Additional comorbidities included arterial hypertension, insulin-dependent diabetes mellitus, permanent pacemaker implantation due to sick-sinus syndrome and reported weight loss of 8 kg since the onset of symptoms.

Due to moderate frailty with a 3.8% STS score and a 1.3% EuroSCORE II she was assigned for minimally invasive SAVR and received a self-expandable SV [PERCEVAL-S®, sizeM, (Livanova, Saluggia, Italy)]. Both the intra- and post-operative course were uneventful. The initial anticoagulation protocol was maintained with low-molecular weight heparin s.c. at a doses of 2× bodyweight in milligram with anti-Xa monitoring and addition of aspirin 100 mg/d, followed by acenocoumarol (INR target 2.0–3.0). Discharge echocardiography showed a mean pressure gradient (MPG) across the AV of 7 mmHg without AV regurgitation. She was readmitted 10 months after the index procedure with symptoms of recurrent heart failure, dyspnoea, pulmonary congestion (oxygen saturation 94%), peripheral oedema, mild end-diastolic murmur, paced heart rate 78 b.p.m., NYHA IV, and increased NT-proBNP.

Diagnostic assessment

Transthoracic echocardiography demonstrated recurrent AV stenosis (AV area 0.92 cm^2), with a MPG of 12 mmHg with severe regurgitation and a preserved LV-EF (*Figure 1A*). Transoesophageal echocardiography (TOE) showed an immobile right-coronary cusp (RCC) due to BVT (*Figure 1B*). Cardiac computed tomography (CT) scan revealed a 12 × 4.9 mm thrombus at the ventricular aspect of the RCC and the right non-coronary commissure (*Figure 2*) with an elliptic expansion shape (22.6 × 18.8 mm) of the bioprosthesis at the annulus level (*Figure 3A*). Any relevant coronary stenosis (>50%) was excluded by angiography prior to SAVR and coronary CT scan prior to ViV-TAVI.

Acquired hypercoagulable disorders or systemic lupus erythematodes were excluded. Neoplastic diseases with paraneoplastic syndrome were excluded in the CT scan, and the patient's initial gastrointestinal symptoms were thoroughly investigated with gastroscopy, colonoscopy, and intrauterine ultrasound.

Interventions

Pharmacological therapy for thrombus resolution was initiated with systemic heparinization (aPTT 50–60 s) followed by acenocoumarol (target INR: 2.5–3.5) for 3 months, according to the current guidelines/recommendations.[4,5] Because of further clinical deterioration and failure of thrombus resolution, the patient was considered for redo-SAVR or ViV-TAVI. She was re-evaluated by the heart-team, which considered the following characteristics: severe frailty with a 10.01% STS score and a 6.9% EuroSCORE II. Due to this high surgical risk, we performed a transfemoral ViV-TAVI under general anaesthesia using a 23 mm Edwards-Sapien 3 bioprosthesis. The lower stent row of the TAVI was positioned under the lowest part of the SV frame to achieve stability of the implanted valve and to engage the thrombus. Intraoperative valve function was excellent showing minimal residual transvalvular gradient (*Figure 4*) without paravalvular regurgitations on TOE, confirmed by aortic root angiography. She was extubated immediately after the procedure with an uneventful post-operative course. She was discharged home on Day 7 after TAVI with the following anticoagulation regime: acenocoumarol (target INR 2–3) and 100 mg aspirin once per day.

Follow-up and outcome

Post-operative echocardiography 5 days, 6, and 12 weeks after TAVI showed regular AV function with a constant minimal MPG (≤10 mmHg) and excluded valvular regurgitation. Computed tomography scan on Day 5 after TAVI confirmed perfect anatomical positioning of the AV with a completely circular shape of the annulus [diameter 20.4 × 20.1 mm (*Figure 3B*)]. The course of NT-proBNP levels was 2260 ng/L before initial aortic valve replacement, 2892 ng/L before

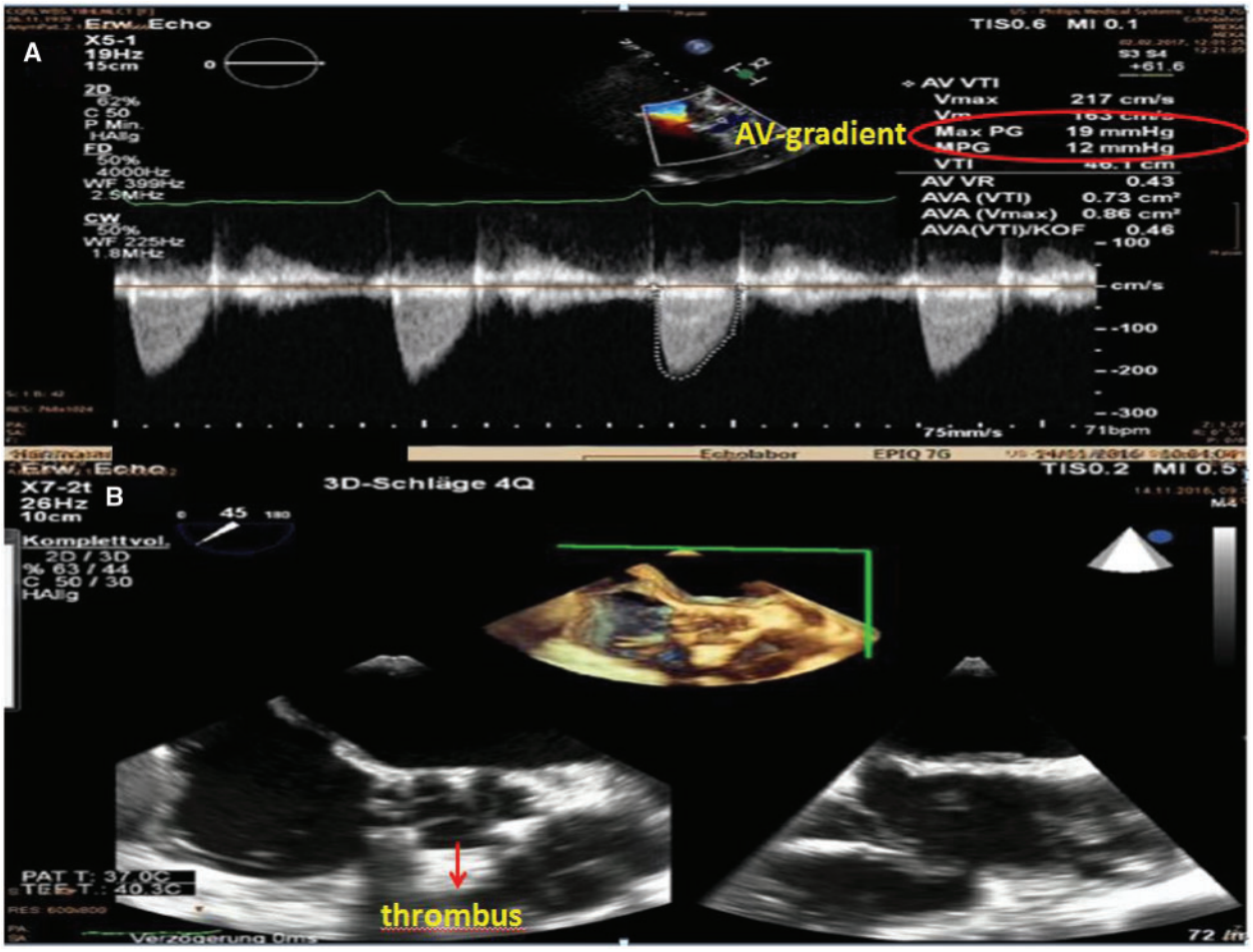

Figure 1 (*A* and *B*) Transoesophageal echocardiography with aortic valve gradient and thrombus pre-valve-in-valve transcatheter valve implantation.

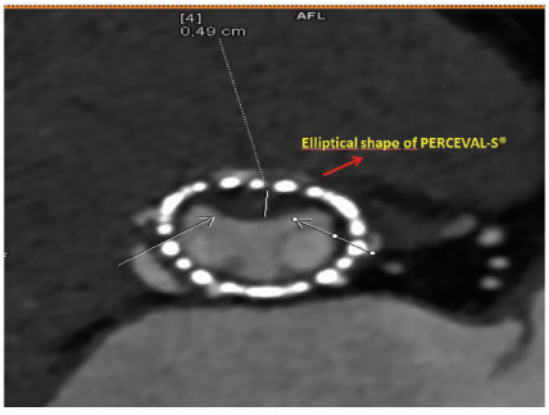

Figure 2 Aortic valve thrombus of PERCEVAL-S® seen by computed tomography scan.

the ViV procedure and 1475 ng/L immediately thereafter (normal range <376 ng/L). The calculated systolic PAP decreased from 40 mmHg before ViV-TAVI to 30 mmHg. The patient presented with NYHA I 1 month after ViV-TAVI.

Discussion

To our knowledge this is the first report of a ViV-TAVI in a patient with a BVT of a self-expandable SV. There are sparse reports of documented SV thrombosis.[6,7] The prevalence of BVT after SAVR or TAVI ranges between 7% and 23%.[8–11] Notably, some studies reported no association with clinical symptoms[9,12] and revealed complete resolution of BVT after vitamin K antagonist treatment. *Egbe et al.*[13] identified a 50% increase in MPG over 5 years as an independent predictor of BVT in 397 patients.

Patient deterioration is probably associated with a combination of stenosis and regurgitation due to BVT as well as with the cardiac

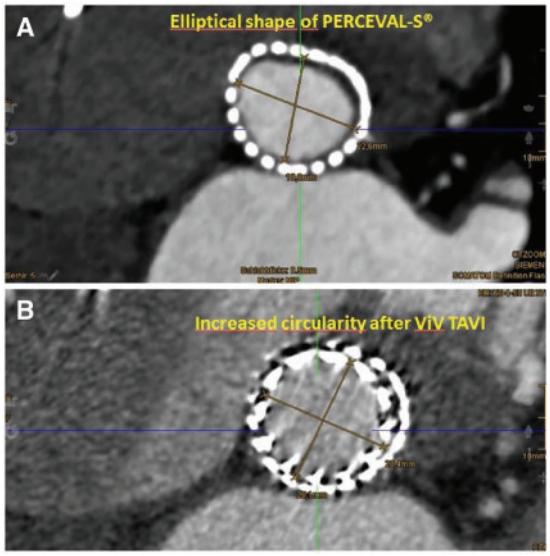

Figure 3 (*A* and *B*) Computed tomography scan pre- (elliptical shape *A*) and post-valve-in-valve transcatheter valve implantation (circular shape *B*).

low-flow condition. The latter is a real challenge in heart failure management with preserved EF and is probably even more challenging when setting continuous right ventricular pacemaker stimulation resulting in a left bundle branch block. In our experience, these patients are extremely difficult to treat, as they have a high perioperative risk and often return with recurrent symptoms.

The evolution of cardiac CT[14] and the advent of three-dimensional TOE have provided appropriate imaging evidence of hypoattenuating leaflet thrombosis, and the reported data are steadily increasing.

There are several technical aspects to be considered when planning and performing ViV-TAVI. First the exact brand, size and position of the index bioprosthesis volume rendering by CT angiography (*Figure 3A*) should be known for precise planning of the new prosthesis type and size. Dysfunctioning SV are considered a special condition, as they are self-expanding and the thrombosis may be associated with the initial anatomical valve position. Remaining calcifications of the native AV or a severely elliptic annulus may prevent proper expansion of the valve and thus pave the way for turbulence with increased shear stress or low-flow phenomena within the SV, which is prone to thrombosis. In our case, the SV was correctly sized and positioned. The form of the PERCEVAL-S® probably makes the use of high-profile TAVI less beneficial due to its difficult positioning (valve trapping or stent engagement) during implantation. We opted for a balloon-expandable valve to increase the circularity of the annulus after the procedure and to avoid tangling with the high frame of the PERCEVAL-S®.

Moreover, the exact position of the thrombus and its mobility should be thoroughly examined due to the risk of embolism inherent in wire manipulations, which should not be underestimated during the procedure.

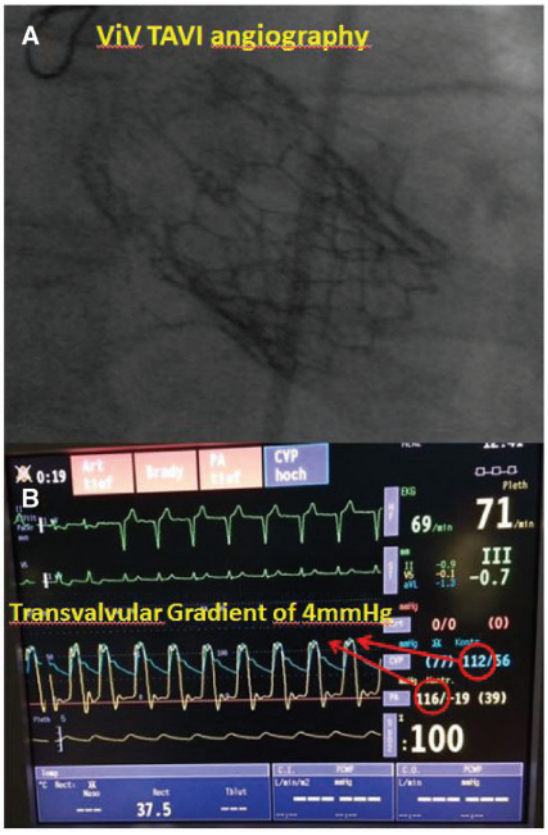

Figure 4 (*A* and *B*) Valve-in-valve transcatheter valve implantation angiography and intra-OP gradient post-transcatheter valve implantation.

Conclusion

To summarize, we present a case of BVT of a SV treated by ViV-TAVI as an alternative to high-risk SAVR with excellent results in the short-term follow-up.[15] However, mid- and long-term results are still outstanding, making this therapy's perspective unclear. This strategy is feasible when specific valve type and anatomical conditions are exhaustively considered.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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