

# Bloody tricuspid stenosis: case report of an uncommon cause of haemoptysis

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## Background

Haemoptysis is usually caused by pulmonary and infectious diseases. In few cases, it has a cardiac cause, such as pulmonary embolism or mitral valve stenosis. Haemoptysis may be an uncommon symptom of prosthetic valve dysfunction, being related to elevated right heart pressures.

## Case summary

A 22-year-old woman from sub-Saharan Africa known for a triple valve replacement was hospitalized for dyspnoea and haemoptysis. A careful clinical evaluation excluded the most common causes of haemoptysis. Transthoracic echocardiogram showed normal biventricular function, normally functioning mechanical prosthetic aortic and mitral valves, and the biological tricuspid prosthesis showed an increased transvalvular gradient. Contrast chest computed tomography scan excluded pulmonary embolism and mechanical valve obstruction, but revealed marked systemic venous hypertension. Right heart catheterization confirmed increased right heart pressures and severe bioprosthetic tricuspid valve stenosis. The patient underwent a successful percutaneous tricuspid valve-in-valve replacement, with complete resolution of symptoms.

## Discussion

The increase in venous pressures due to bioprosthetic tricuspid stenosis caused veno-venous shunts: blood from the lower body was drained into the superior vena cava via the azygos vein. Increased pressure in the latter affected pressure in bronchial veins and arteries, leading to haemoptysis. Cardiac surgical reinterventions are associated with worse outcomes and higher mortality rates. Management of a degenerated prosthetic tricuspid valve is challenging and requires a multidisciplinary assessment. Transcatheter tricuspid valve replacement is becoming a feasible option in patients with prosthetic dysfunction. Based on evidence to date, tricuspid valve-in-valve replacement appears to be a safe, feasible, and effective alternative in selected young patients.

## Keywords

Haemoptysis • Venous hypertension • Prosthetic valve • Tricuspid valve stenosis • Transcatheter valve replacement • Case report

## Learning points

- Tricuspid stenosis could be a cause of haemoptysis.
- Highlight the symptoms of venous hypertension secondary to tricuspid valve stenosis.
- Percutaneous interventions could be an option for patients with a degenerated tricuspid valve prosthesis.

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## Introduction

Nowadays in Europe haemoptysis is uncommonly caused by heart valve disease, being classically related to rheumatic mitral stenosis.<sup>1</sup> Indeed, tricuspid prosthetic valve stenosis presenting with haemoptysis is a rare finding. In the present paper, we will present a case of a degenerated bioprosthetic tricuspid valve causing haemoptysis in a young patient, with a focus on its challenging management strategies.<sup>2</sup>

## Timeline

Six years before	Triple valve replacement for rheumatic heart disease
Five years before	Stroke with subsequent recovery
Three weeks before	Haemoptysis onset
One week before	Dyspnoea onset and worsening of haemoptysis
Arrival at our hospital	Hospitalization
Day 1	Echocardiogram showed normally functioning aortic and mitral prosthetic valve and an increased transvalvular gradient over the tricuspid bioprosthesis
Day 2	Contrast chest computed tomography scan showed systemic venous hypertension
Day 16	Right heart catheterization showed increased right heart pressures and confirmed the severe bioprosthetic tricuspid valve stenosis
Day 32	Percutaneous tricuspid valve-in-valve replacement
Day 34	Post-procedural echocardiogram
Day 36	Hospital discharge
Day 195	Follow-up clinical assessment and echocardiogram, showing

## Case presentation

A 22-year-old woman from sub-Saharan Africa presented to our emergency department after 1 week of dyspnoea, leg swelling, and haemoptysis. Haemoptysis had started as blood-streaked sputum 3–4 weeks before, gradually increasing to multiple daily blood clot expectorations within 2 weeks.

In her past medical history, we highlight a triple valve replacement for rheumatic heart disease at age 16 (19-mm CarboMedics mechanical aortic valve prosthesis; 27-mm CarboMedics mechanical mitral valve prosthesis; 27-mm Magna biological tricuspid valve prosthesis). Her medical history also comprised a right middle cerebral artery stroke at age 17, conditioning left hemiplegia and aphasia at presentation with subsequent full recovery. In addition, she was known for chronic haemolytic anaemia and thalassaemia trait. The patient

denied any allergy, and her family history was unremarkable. She was taking vitamin K antagonists for anticoagulation.

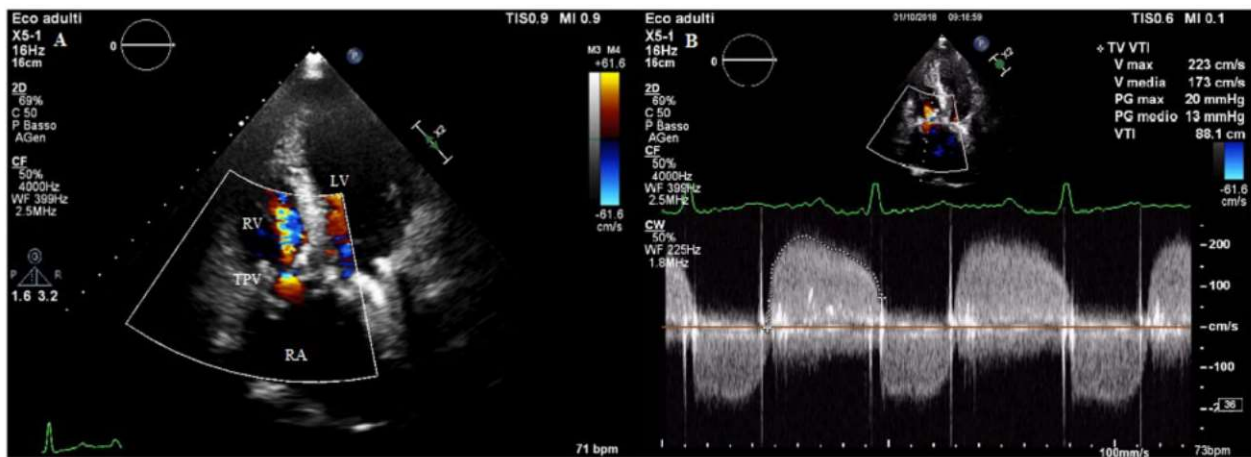
Upon presentation, the patient was afebrile and her blood pressure was 120/70 mmHg. She was comfortable at rest, New York Heart Association (NYHA) class III. Heart rate was 110 b.p.m. and oxygen saturation was 99% on room air. Her physical examination was characterized by prosthetic heart sounds with a 4/6 systolic murmur, diminished breath sounds in lung bases, mild hepatomegaly, moderate bilateral ankle oedema, and mild jugular venous distention.

Electrocardiogram on presentation showed sinus tachycardia at 105 b.p.m. with first-degree atrioventricular block. Arterial blood gas test showed compensated respiratory alkalosis. Lab tests showed mild microcytic anaemia (haemoglobin 11.8 g/dL, reference interval (RI) 12.5–14.0 g/dL) and an International Normalized Ratio of 4.13 (RI 0.80–1.16). Renal function and electrolytes were between normal ranges, while mild liver enzyme alterations were recorded (total bilirubin 3.8 mg/dL, RI 0.20–0.90 mg/dL). Brain natriuretic peptide was 401 pg/mL (RI 10–100 pg/mL). C-reactive protein and procalcitonine were both negative. Chest X-ray showed an enlarged heart with minimal bilateral pleural effusion.

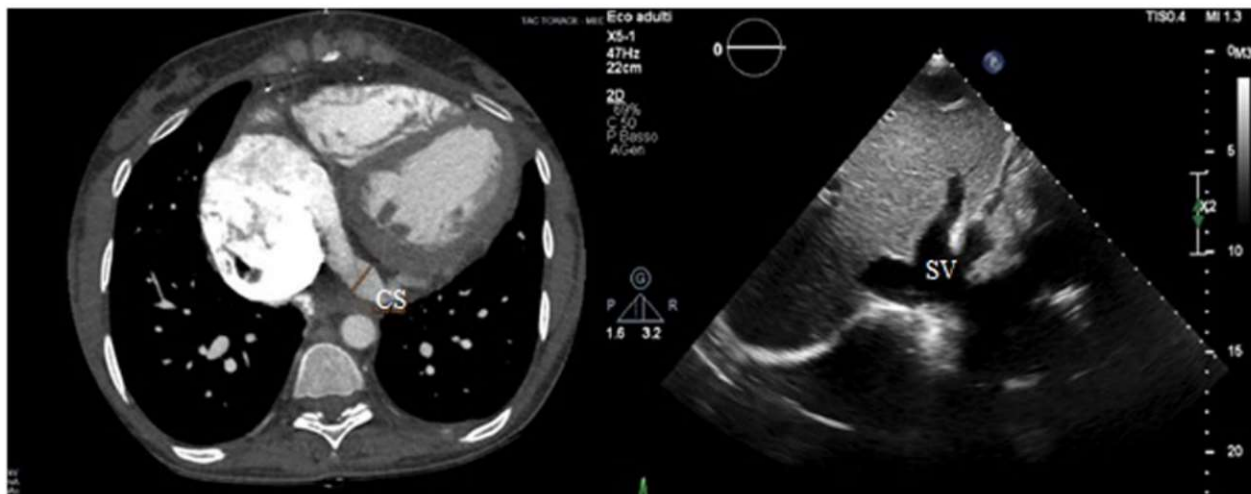
The patient was hospitalized for heart failure and haemoptysis. Daily treatment with intravenous furosemide 40 mg and bisoprolol 1.25 mg was started. Haemoptysis was treated with inhaled tranexamic acid 500 mg twice a day and iced saline lavages. Warfarin was stopped; enoxaparin 6000 IU twice a day was started. Airway bleeding rapidly improved.

Chest computed tomography (CT) scans excluded pneumonia, bronchiectasis, and neoplasms.<sup>3</sup> Pulmonary tuberculosis was ruled out with a negative sputum smear examination and negative molecular tests for *Mycobacterium tuberculosis*. Echocardiogram during sinus tachycardia showed normally functioning prosthetic aortic and mitral valves, a hyperdynamic left ventricular wall motion with above-normal ejection fraction. Right ventricular longitudinal function was preserved and the biological tricuspid prosthesis showed an increased transvalvular gradient (mean gradient 13 mmHg, *Figure 1*) and a mild-to-moderate regurgitation; pulmonary artery systolic pressure could not be estimated. Inferior vena cava (IVC) was dilated (22 mm) with reduced collapsibility.<sup>4</sup> No echocardiographic finding was suspicious for infective endocarditis. Contrast chest CT scan excluded pulmonary embolism, arteriovenous malformations, and additional scans excluded possible mechanical valve obstruction, such as pannus or thrombi.<sup>5</sup> It also revealed marked systemic venous hypertension, manifesting as dilation of venae cavae, suprahepatic veins, cardiac veins, and of the coronary sinus (*Figure 2A and B*). It showed also veno-venous shunts, as well as mild ascites and hepatomegaly. Right heart catheterization confirmed increased right heart pressures, severe bioprosthetic tricuspid valve stenosis (mean gradient 9 mmHg), elevated venous pressure in venae cavae, and a mean right atrial pressure of 22 mmHg (*Figure 3*). The increase in venous pressures due to tricuspid stenosis caused veno-venous shunts, resulting in augmented venous pressure in the azygos vein and in the bronchial circulation, eventually leading to haemoptysis.

Severe symptomatic tricuspid stenosis was referred for surgery.<sup>2</sup> Due to the high operative mortality and morbidity of redo tricuspid valve surgery,<sup>6</sup> pre-operative evaluation was challenging. Eventually, due to the patient's age and the possible long-term need for reoperation,



**Figure 1** (A) Doppler appearance of stenosis jet on echocardiogram. LV, left ventricle; RA, right atrium; RV, right ventricle; TPV, tricuspid prosthetic valve. (B) Transtricuspid pressure gradients derived using the Bernoulli equation: peak maximum velocity 223 cm/s; mean gradient 13 mmHg.



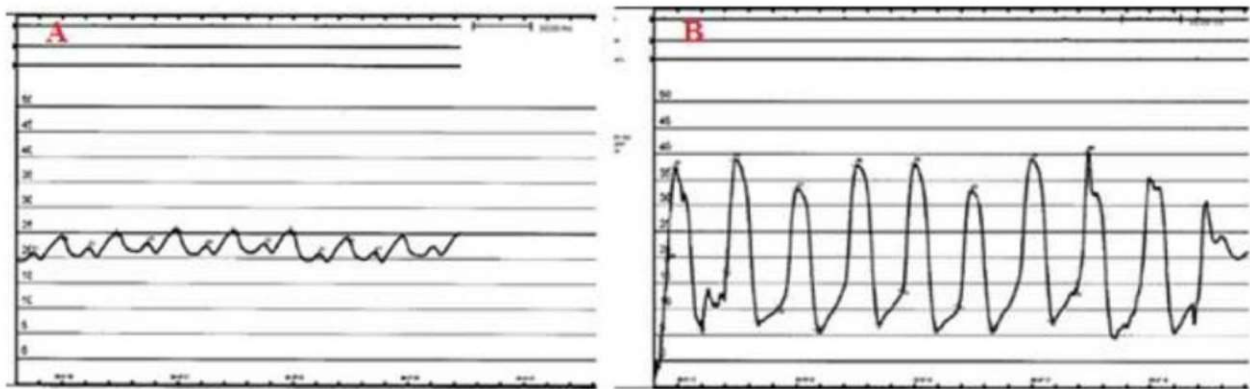
**Figure 2** (A) Coronary sinus (CS) dilation on contrast computed tomography scan. (B) Suprahepatic veins dilation on echocardiogram (SV).

we opted for a percutaneous approach. The patient underwent transfemoral tricuspid valve-in-valve replacement, using an Edward Sapien 3 (29 mm) bioprosthesis (Figure 4A). Early post-procedural echocardiogram showed a normally functioning bioprosthetic tricuspid valve, no paravalvular leakage, a mean transvalvular gradient of 4–5 mmHg (Figure 4B), and a slight reduction in IVC diameter (19 mm).

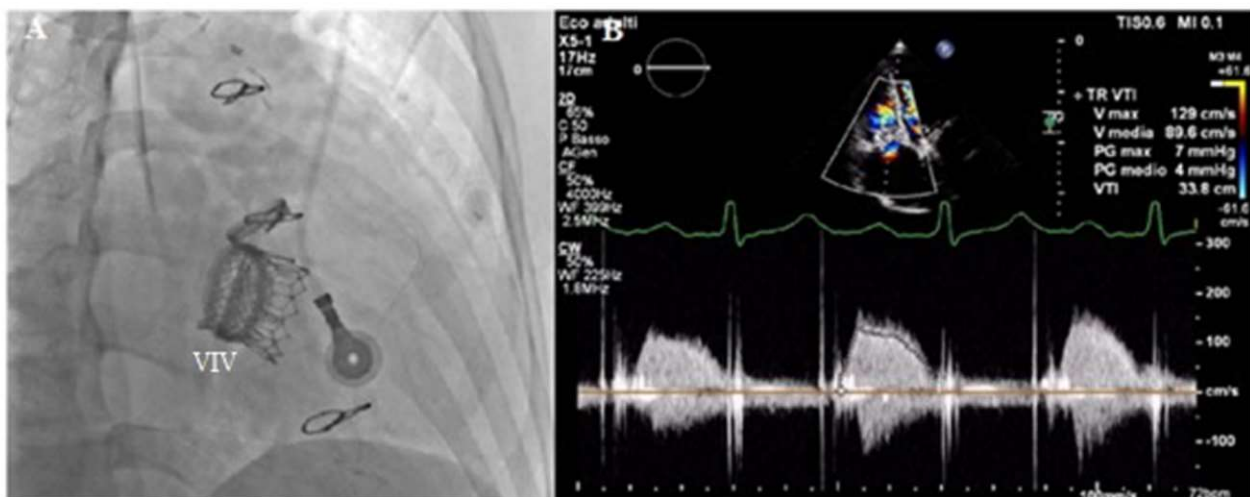
The patient was discharged asymptomatic in NYHA class I. Discharge medications included bisoprolol 1.25 mg, lansoprazole 30 mg, and warfarin. Complete resolution of airway bleeding was recorded. During a follow-up visit at 6 months, the patient denied any airway bleeding but complained of occasional palpitation. Echocardiogram at 6 months showed normally functioning tricuspid valve-in-valve bioprosthesis, mean transvalvular gradient 5 mmHg, no paravalvular leak; IVC diameter 12 mm.

## Questions

- (1) Haemoptysis ranges from blood-streaked sputum to the presence of blood in bronchi. Pulmonary diseases are its most common cause, but there are a few peculiar cardiac conditions which may lead to haemoptysis. Which of the following statements is false?
  - a. Pulmonary veins isolation may be complicated by haemoptysis.
  - b. Less than 2% of patients with pulmonary embolism present with haemoptysis.
  - c. Mitral stenosis may lead to haemoptysis.
  - d. Endocarditis may cause haemoptysis.



**Figure 3** Right heart catheterization. (A) Right atrium pressure curves (mean pressure 23 mmHg). (B) Right ventricle pressure curves (systolic/diastolic/mean pressures: 37/7/11 mmHg).



**Figure 4** (A) Fluoroscopic image after the tricuspid valve-in-valve replacement (VIV). (B) Post-procedural echocardiogram with a significant decrease in pressure gradients, with a peak maximum velocity of 129 cm/s and a mean gradient of 4 mmHg.

- e. Tricuspid stenosis may be associated with haemoptysis.
- (2) Which of the following is the least specific of haemodynamically significant tricuspid stenosis?
- Mean pressure gradient >5 mmHg.
  - More than moderate right atrium dilation.
  - Valve area by continuity equation <1 cm.
  - Inflow time-velocity integral >60 cm.
- (3) While the potential role of transcatheter tricuspid valve interventions still needs to be determined, transcatheter aortic valve implantation procedures are routinely used—especially in elderly patients. Which of the following statements regarding transcatheter valvular interventions is false?
- Transcatheter valve implantations in the mitral position are not feasible in symptomatic elderly patients who are inoperable.

- The potential role of transcatheter tricuspid valve treatment in high-risk patients needs to be determined.
- Oral anticoagulation is recommended for patients with transcatheter-implemented bioprostheses who have other indications for anticoagulation.
- Before transcatheter valve procedures, it is essential to evaluate the feasibility of various access routes.

## Discussion

There are numerous causes of haemoptysis: pulmonary diseases, vascular diseases, infections, neoplasms, bleeding disorders, drugs, and toxins use.<sup>7</sup> Bronchoscopy and chest CT scans usually help in the suspicion of airway of parenchymal diseases. Moreover, laboratory tests and medical history might exclude bleeding disorders and substance abuse.

Heart valve diseases are a potential, albeit infrequent, cause of haemoptysis. In these cases, haemoptysis is due to an increased post-capillary pressure, which is usually encountered in patients with severe mitral stenosis and advanced heart failure. Haemoptysis secondary to biological tricuspid prosthesis stenosis, to the best of our knowledge, has never been described in medical literature. The pathophysiology is, however, rather simple. The increase in venous pressures due to tricuspid bioprosthesis stenosis caused venovenous shunts: blood from the lower body was drained into the superior vena cava via the azygos vein. Increased pressure in the latter affected pressure in bronchial veins and arteries, leading to haemoptysis.

Management of tricuspid valve disorders and of degenerated bioprosthetic tricuspid valves in young patients is complex. Biological prostheses are usually preferred due to their adequate long-term durability and lower risk of thrombosis in the tricuspid position. In spite of this, reoperation rate ranges between 10% and 20% at 9 years, and ~4 in 10 patients will require a redo procedure within 15 years. This trend is even more evident in younger patients: below 16 years of age, the rate of reoperation, and dysfunction at 5 years is 30% and 70%, respectively.<sup>8–10</sup> The potential number of open-heart surgeries that young patients with valve diseases may require during the course of their life warrant consideration of other strategies.

Although at an early stage, transcatheter tricuspid valve replacement has become a feasible option in patients with prosthetic dysfunction and high surgical risk, or patients with a history of several tricuspid interventions.<sup>11–13</sup> Transcatheter implantation of valves into degenerated tricuspid bioprostheses (valve-in-valve) has emerged as an alternative to open surgery. CoreValve (Medtronic, Minneapolis, MN, USA), SAPIEN (Edwards Lifesciences, Irvine, CA, USA), and Melody valve (Medtronic, Minneapolis, MN, USA) have all been approved by FDA for such indication.<sup>14,15</sup>

## Conclusions

Management of a degenerated prosthetic valve in patients with multiple valve disease is challenging and requires a multidisciplinary assessment. Based on the available evidence to date, tricuspid valve-in-valve replacement appears to be a safe, feasible, and effective alternative to redo in selected young patients.

## Lead author biography



Filippo Trombara is a cardiology resident from the University of Milan. He graduated from the University of Modena in 2016. After that, he moved to Milan to attend cardiology training at Monzino Cardiology Center IRCCS, where he is developing interest in acute cardiac care.

## 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 authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

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