

### Aortomitral Curtain Abscess Due to *Actinomyces Denticolens*, a Rare Entity of Infective Endocarditis: A Case Report

The incidence of infective endocarditis (IE) has increased in recent decades, with a reported 30-day mortality of up to 30%.

Over the past three decades, more than 25 *Actinomyces* species have been identified in humans. Laboratory detection and recognition as pathogens in clinical settings can be challenging. (1) It is a Gram-positive bacillus. The first microorganism of this family was described in 1896, but *Actinomyces denticolens* was reported by I. Batty in 1958. Its main reservoir is the human being, and it is found mostly in the oral cavity at a very early age; it has also been described in the pharynx and the esophagus. (2)

Actinomycosis is a rare, chronic, slowly progressive granulomatous disease. It is often misdiagnosed because it can closely mimic tuberculosis and even malignant diseases. (3) Prevalence rates are unavailable. It is classified into different clinical forms according to the infected or disseminated body site, *A. israelii* being the major causative agent in most presentations. (3)

So far, 26 cases of *Actinomyces* sp. as a cause of IE have been reported in the literature, with isolation of *A. neuii*, *A. funkei*, *A. israelii*, *A. viscosus*, and *A. meyeri*.

*Actinomyces denticolens* has not been described as a cause of IE; therefore, this is the first case reported in the literature.

A 39-year-old male patient, a farmer from a rural area, with no relevant medical history, presented with a 3-month history of dyspnea, fever, and weight loss. Initially, the patient consulted in another medical center, where he was diagnosed with bilateral pleural effusion and was managed with percutaneous drainage. The complication was hemodynamic and respiratory collapse, requiring vasopressor and ventilatory support. Transthoracic echocardiography revealed severe aortic regurgitation with eccentric hypertrophy and left ventricular ejection fraction (LVEF) of 37%. After achieving hemodynamic stability and weaning off inotropes, the patient was transferred to our institution for comprehensive, tertiary care management.

Physical examination on admission revealed carotid dance and diastolic murmur in the aortic area, bounding pulses, bibasilar inspiratory rales, use of accessory muscles, and anasarca.

Transesophageal echocardiography showed left atrial enlargement, intracavitary thrombus, LV enlargement with eccentric hypertrophy (end-diastolic volume 383 ml by biplane method), and LVEF 30%. Vegetation on the atrial side of the mitral valve was observed, with moderate functional regurgitation. Vegetation attached to both bicuspid aortic valve leaf-

lets (the largest, 23 mm), severe regurgitation, pressure half time of 114 m/s. Enlargement of the right chambers with preserved contractility. Systolic blood pressure: 69 mm Hg. Blood cultures were negative.

The patient was transferred to surgery. Sievers type I bicuspid aortic valve was confirmed by fusion of the calcified coronary leaflets in the facet and partial rupture of the raphe close to the coaptation point. Lumpy vegetations with thrombus formation on the free edge were evident in the ventricular aspect. A pocket involving the anterior mitral valve leaflet was observed at the aortomitral junction, with vegetations and purulent secretion (Figure 1). Removal of the infectious material revealed a perforation of the anterior mitral valve leaflet, also involving the non-coronary aortic valve leaflet. The aortic valve was resected, the mitral valve was repaired, and an autologous pericardial patch was deployed to close the perforation (Figure 2). Aortic valve replacement with mechanical prosthesis and exclusion of the left atrial appendage was performed. Intraoperative echocardiography demonstrated a well-positioned aortic valve prosthesis, and absence of mitral regurgitation.

Postoperative course was favorable, and the patient was weaned from mechanical ventilation within a few hours after surgery, requiring milrinone for 3 days. Control echocardiography showed no evidence of residual vegetations, confirming the intraoperative echocardiography findings. The aortic valve culture evidences *Actinomyces denticolens* growth. Intravenous antibiotic management is adjusted with penicillin and doxycycline for 6 weeks, followed by oral



Fig. 1.

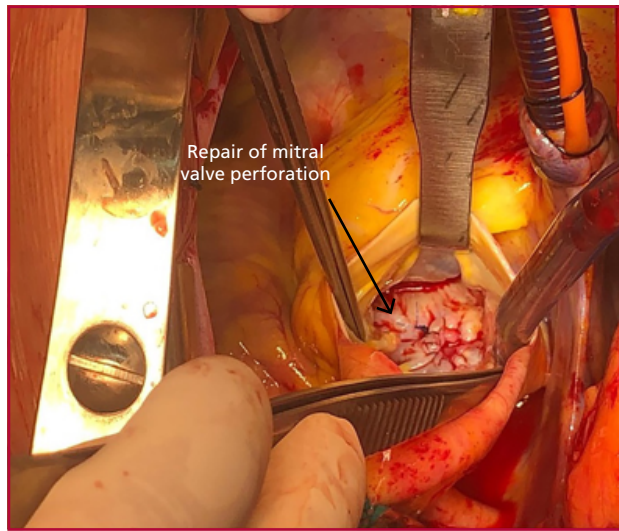


Fig. 2.

oxacillin and amoxicillin, completing 6 months as recommended by Infectology.

Currently, the patient is in good general condition, anticoagulated with warfarin in therapeutic range.

Left-sided native valve IE remains the most common presentation of IE, accounting for 70% of all cases. (5) Bicuspidization of the aortic valve is the main predisposing factor, as in our case. The treatment for this patient created surgical challenges as a result of the delayed consultation due to social conditions. When the patient finally consulted, he was already in heart failure and pulmonary edema, requiring intensive care management due to severe deterioration of the left ventricular function.

In cases of IE of the aortic valve, there is lack of evidence on superiority of repair over replacement. (4) In our patient, extensive aortic valve destruction did not allow for any approach other than replacement.

The decision on what type of prosthetic valve to choose is based on the usual criteria. (5) However, some considerations must be taken into account in patients with IE, including current neurological involvement, prolonged anticoagulation, and the risk of cerebral bleeding events. In these cases, since anticoagulation could cause major complications, bioprosthesis implantation is a better option. Mechanical aortic valve replacement was performed in our patient given his age, the presence of intracavitary thrombus, and the integrity of his neurological status.

In cases of native mitral valve IE, clinical practice guidelines suggest repair whenever possible, when there are no valvular abscesses or severe involvement of the subvalvular apparatus. (5, 6)

In our patient, the mitral valve disease was an extension of the aortic infection. Mitral valve repair was preferred due to well-defined perforation, strong edges, and favorable surgical exposure. Cleansing of the outflow tract was achieved, and the well-defined

abscess allowed for fine debridement of its edges and placement of a pericardial patch.

This is the first case of subacute infective endocarditis due to *Actinomyces denticolens* reported in the literature, which should be considered in culture-negative endocarditis. In addition, successful surgical management of a complex case is presented, which may be a reference for future cases.

#### Conflicts of interest

None declared.

(See authors' conflicts of interest forms on the website/ Supplementary material).

#### Ethical considerations

Not applicable.

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#### Percutaneous Closure of Complex Atrial Septal Defect: Unusual Implantation of Two Devices

Persistent patent foramen ovale (PFO) in adults is a common finding with a prevalence of 25% in the general population, with no clinical implications in most cases. Between 15-35% of ischemic strokes are classi-

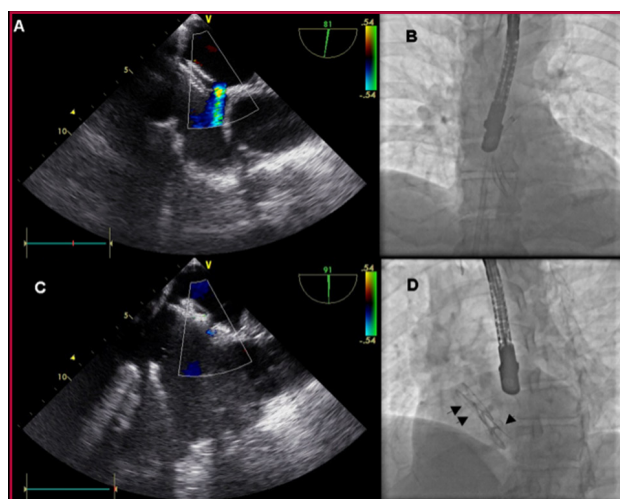
fied as cryptogenic, despite an exhaustive research for potential etiologies. Likewise, half the patients < 60 years of age with cryptogenic stroke have PFO. There is evidence that accounts for percutaneous PFO closure to avoid recurrence in this clinical setting.

The fossa ovalis may have more than one defect of different sizes and locations, and the presence of residual leak after percutaneous closure increases the risk of stroke recurrence or transient ischemic attack.

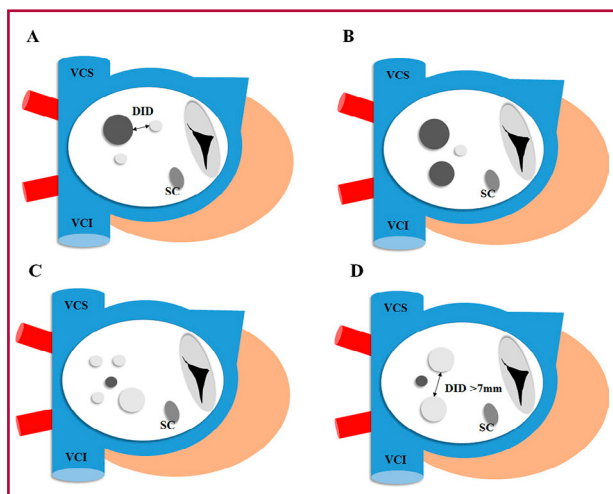
We report the case of a 51-year-old male patient with acute onset of left hemiparesis associated with expressive aphasia, lasting less than 24 hours. A magnetic resonance imaging of the brain showed a right postcentral parietal cortex lesion with posterior insular involvement. This lesion had T2 Flair hyperintensity with restricted diffusion. Vital signs were within normal limits on admission. ECG detected sinus rhythm, and the patient was started on aspirin and statins. Blood tests ruled out hypercoagulability syndrome. No atrial flutter or atrial fibrillation was observed on 24-hour Holter monitoring. Atherosclerotic plaques in carotid arteries were ruled out, and a transesophageal echocardiography (TEE) ruled out aortic plaques or thrombi in the left atrial appendage. The interatrial septum was hypermobile and suggestive of atrial septal aneurysm.

Agitated saline test showed more than 20 bubbles passing from the right to the left atrium. A RoPE score of 7 was indicative of percutaneous PFO closure.

Transesophageal echocardiography-guided percutaneous intervention revealed a large hypomobile



**Fig. 1.** (A) Transesophageal echocardiography view (90° plane) with slight cranial angulation. The image shows a released patent foramen ovale device with no proper occlusion of the residual atrial septal defect. (B) Fluoroscopy image showing the release of the patent foramen ovale device and the sheath through the anterosuperior residual defect. (C) Transesophageal echocardiography image showing both devices with absence of residual shunts. (D) Fluoroscopy image of both devices released with the 'sandwich' technique. Atrial septal defect device (double arrow) embracing the patent foramen ovale device (single arrow).



**Fig. 1.** Classification of multi-hole defects. Type A: a big defect associated with one or more small defects, with different inter-defects distances (IDD). Type B: a big defect and a moderate defect, associated with one or more small holes with different IDD. Type C: cribriform septum with 5 or more defects. Type D: small and moderate defects with  $IDD \geq 7$  mm. Darkest defects should be chosen for device implantation. Large defect ( $\geq 15$  mm), moderate defect (5-14 mm) and small defect (< 5 mm). Coronary sinus (CS). Adapted from Farhaj Z et al (3).

atrial septal aneurysm with a large distended PFO (3 mm), and another 5 mm-diameter small defect close to the PFO. A Nit Occlud® PFO device (pfm Medical, Cologne, Germany) with a disc diameter of 30 mm was implanted, which was observed—prior to be released—to cover the additional defect. However, the anterosuperior atrial septal defect (ASD), of moderate size (12 mm in diameter), persisted after the release (Figure 1A).

Since persistent residual shunts could not be tolerated, a catheter was advanced through the residual defect (Figure 1B) and a Nit Occlud ASD-R device (pfm Medical, Cologne, Germany) with a diameter of 18 mm was implanted. The second device was embracing the first device, achieving complete occlusion of both defects (Figure 1C). The patient was discharged without complications on the following postoperative day. At 9-month follow-up, no recurrent events or arrhythmias were observed, and both devices were in normal position (Figure 1D).

In patients with more than one defect, percutaneous intervention techniques vary and include the implantation of a single oversized device to cover both defects, the interatrial septum rupture with a balloon unifying both defects, the use of devices with small waist multi-hole ASD or PFO type, and finally the implantation of two devices using interlocking or sandwich technique when the distance between the defects is > 7 mm. (1)

Masseli et al. (2) propose a single device implantation to occlude both defects since, in their published series, the rate for complete closure of both defects

was 26% at 1 month, and increased to 78% at 24 months.

A recent classification of multi-hole defects according to size, number and inter-defects distance indicates that type A defects can be treated with an ASD device; for type B, it is suggested to try an ASD device and evaluate the need for a second device, and types C and D should be approached with a PFO device to occlude the middle hole (Figure 2). (3)

When the implantation of two devices is required, two femoral venous access points are necessary. The devices should be implanted simultaneously, first the smallest and then the largest, and be released in the same order. When multiple devices are used, the procedure is technically challenging and requires an expert operator. Moreover, the risk of device embolization and erosion is higher than in single defects. (4)

The risk for a residual shunt after percutaneous PFO closure is four times higher for recurrent thromboembolic events, compared with patients without residual shunts; (5) therefore, a second device should be considered in patients with significant residual shunts. (6)

This case represents an uncommon approach due to the need of an additional ASD device implantation after successful occlusion of a PFO with a suitable device, in order to prevent residual shunts in the context of secondary prevention of PFO.

#### Conflicts of interest

None declared.

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#### Ethical considerations

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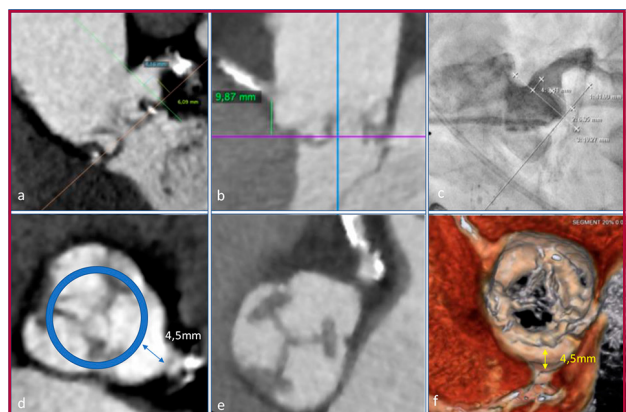
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### Intentional Laceration of the Aortic Valve Bioprosthesis to Prevent Coronary Occlusion During Transcatheter Aortic Valve Implantation

Coronary occlusion is a rare complication with severe consequences during transcatheter aortic valve implantation (TAVI), which usually occurs in about 1% of cases; (1) this complication increases 4-6 times during a Valve-in-Valve (ViV) procedure, in which a percutaneous valve is implanted inside a failed implanted bioprosthetic valve, (2) especially when the height of the coronary ostium is < 10 mm compared to the annulus or the distance between the surgical valve and the ostium is < 4 mm. The risk is also increased in valves with leaflets mounted outside of the annulus, and in cases where the orientation of the aorta suggests that the TAVI valve will be closer to a coronary ostium. (3)

To prevent this complication, stents protruding into the aorta outside the TAVI valve have been used with very positive outcomes in the acute stage; however, the difficulty in reaccessing the vessel if needed is a major limitation, especially in young patients who are more likely to require it.

To reduce the risk of this complication and avoid this limitation, the "BASILICA" (Bioprosthetic Aortic Scallop Intentional Laceration to Prevent Iatrogenic Coronary Artery Obstruction) technique was devel-



**Fig. 1.** a & c. Height and distance measurements from the previous valve to the left coronary artery (LCA) ostium. b. Height of the right coronary artery ostium. d. Virtual distance to the LCA ostium. e. Relationship between the LCA ostium and the previous valve posts. f. Distance to the LCA ostium.

oped, consisting of perforation and laceration of the bioprosthetic leaflet with a coronary guidewire. For this purpose, a 0.014" coronary guidewire is used to perforate the leaflet, stimulated by an electrosurgical scalpel; the guidewire is advanced to the left ventricular outflow tract where it is looped and then removed by means of the electrosurgical scalpel stimulation; this contact causes an iatrogenic leaflet laceration to reduce the risk for coronary occlusion by leaving the leaflet laceration in front of the coronary ostium. If necessary, it can be used for one or both ostia.

Our case is a 76-year-old male patient with a history of hypertension, dyslipidemia, non-insulin diabetes, and prostate cancer for which he had received radiotherapy. Thirteen years earlier, the patient had undergone aortic valve replacement with the implantation of a No. 25 bovine pericardial bioprosthesis (Epic, St Jude Medical No 25), mammary artery bypass graft to the anterior descending artery, venous bypass graft to the right coronary artery, and a sequential venous bypass graft to the circumflex-circumflex artery. Four years after implantation, the patient developed infective endocarditis, which was resolved with antibiotics.

In recent months, the patient referred dyspnea progressing to NYHA functional class III prior to current hospital admission. Doppler echocardiography revealed left ventricular (LV) ejection fraction of 55%, preserved ventricular diameters, apical and inferoapical septal hypokinesis, mild left atrial enlargement, and prosthetic dysfunction with a mean gradient of 41 mmHg and an area of 0.82 cm<sup>2</sup>.

Coronary angiography showed proximal occlusion of non-dominant right coronary artery; left main coronary artery without lesions; occlusion of the proximal segment of the anterior descending artery; occlusion of the proximal segment of the circumflex artery; left mammary artery bypass graft to the anterior descending artery without lesions; aorto-coronary venous by-

pass to the first diagonal artery, and sequential bypass graft of the first marginal - posterior descending circumflex artery without lesions.

STS score for mortality was 7.6%, and creatinine clearance was 54.2 ml/min/1.73 m<sup>2</sup>. Upon evaluation, our hospital Heart Team suggested percutaneous aortic valve replacement.

A CT angiography prior to TAVI showed that the height of the left coronary artery ostium was < 10 mm, and the estimated distance between the valve and the ostium was 4.15 mm; therefore, it was a case of intermediate risk because a large ostium was in contrast to those distances (Figure 1). Given that — as explained above— stents are not recommended in relatively young patients, the BASILICA technique prior to TAVI was the option, in order to protect the left coronary ostium.

A right femoral arterial puncture was performed under conscious sedation, and the Pre-Close technique was used with two ProGlide sutures. In addition, a contralateral femoral puncture was performed, a protection 0.018" x 300 cm guidewire was placed for subsequent percutaneous closure, and an 8F sheath was placed in the right femoral vein through which a transient pacemaker lead was introduced.

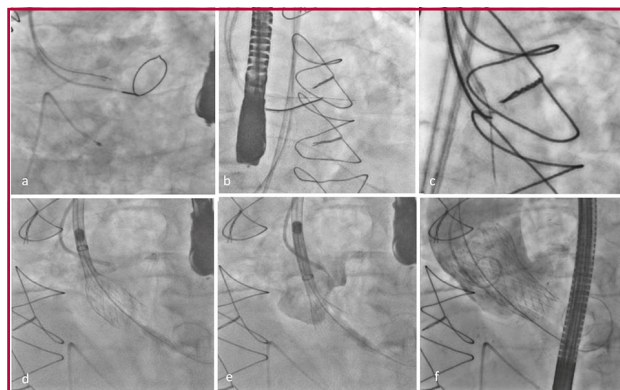
Under fluoroscopic guidance and transesophageal echocardiography, an AL 2 6F Amplatz curve guiding catheter was advanced, and a 130 cm right coronary artery diagnostic catheter (JR3.5) was advanced inside the catheter to the base of the outer wall of the left bioprosthetic leaflet; then a 20-mm loop catheter was positioned in the left ventricular outflow tract (LVOT). A high-support coronary guidewire was advanced and connected at the distal end to the electrosurgical scalpel outside the patient's body, and the leaflet perforation was made using a 40W cutting technique. After passing through the leaflet and once in the LVOT, it was looped. Once both catheters were connected, leaving about 2 cm exposure of the guidewire that was in contact with the leaflet, the coronary guidewire was stimulated with the electrosurgical scalpel with a cutting power of 70W; simultaneously, the looped guidewire was withdrawn towards the aorta, gently pulling with both catheters and causing the laceration of the left bioprosthetic leaflet.

Then, catheterization of the left coronary artery with a guide 6F XB was performed; a 0.014" guidewire was advanced into the anterior descending artery to protect the ostium in case of insufficient laceration, and then an Evolut PRO valve N° 34 was implanted (Figure 2).

Absence of aortic regurgitation and a mean residual gradient of 8.5 mmHg were confirmed after implantation.

The patient was transferred to the coronary care unit, made good progress and was discharged after 24 hours.

Factors contributing to coronary artery occlusion include leaflet characteristics (calcifications, length,



**Fig. 1.** a. Prosthetic valve leaflet perforation. b. Guidewire looping into the ventricular chamber. c. Leaflet cutting with electrosurgical scalpel connected to the guidewire. d & e. Positioning of self-expandable aortic valve by catheterization (TAVI). f. Final outcome showing patency of the left coronary ostium and correct TAVI implantation

thickness, implantation outside the prosthetic valve annulus), small sinus of Valsalva, distance from the annulus to the coronary ostium < 10 mm, estimated distance between the prosthetic valve and the ostium, changes in sinus dimensions (systole versus diastole), type of bioprosthesis, and characteristics of the percutaneous valve. (4)

In his first publication on 7 patients, Khan demonstrated that this leaflet laceration strategy is feasible, which was successfully performed in both coronary arteries without major complications at 30 days. (5) Furthermore, this author demonstrated that the technique appears feasible in a larger series of 30 patients with 37 lacerations: 35 were successful and only 2 presented significant hemodynamic compromise, which were immediately resolved. There was one death and one disabling stroke at 30 days. (6)

This technique requires a significant number of resources and training—not only of the interventional cardiologists, but also of the echocardiographer and the anesthesiologist—to guide the procedure and act efficiently and in coordination in the event of hemodynamic decompensation due to severe aortic regurgitation caused by laceration.

In brief, we report our first case and, to our knowledge, one of the first cases performed in Latin America with this technique that protects the coronary ostium in cases of TAVI for ViV procedures and unfavorable anatomy, as was the case in our patient.

#### Conflicts of interest

None declared.

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#### Ethical considerations

Not applicable

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### Surgical Treatment of Carcinoid Heart Disease: Case Reports

Neuroendocrine tumors are rare malignancies, with an incidence ranging from 2.5 to 5 cases per 100,000 inhabitants. They can occur anywhere in the body but most often originate in the gastrointestinal tract. Carcinoid tumors usually grow slowly, over years, commonly clinically silent, until they have metastasized. They frequently metastasize to regional lymph nodes, to the liver and, less frequently, to the bones. The risk of metastasis is associated with the size of the tumor. The incidence of metastases is less than 15% with a carcinoid tumor smaller than 1 cm but rises to 95% with tumors larger than 2 cm. These tumors may be symptomatic only episodically, and their existence may go unrecognized for many years. Approximately 30% to 40% of patients (mainly with midgut carcinoids) present with features of carcinoid syndrome, manifested by episodes of vasomotor changes (flushing and hypotension; less frequently, hypertension), diarrhea and bronchospasm, or develop these symptoms during the course of the disease. (1)

Cardiac valve involvement associated with carcinoid tumors is a rare entity present in up to 60% of patients with metastatic carcinoid tumors. The pathophysiology of carcinoid heart disease is poorly understood; however, chronic exposure to excessive circulating serotonin is considered one of the most important contributing factors.

Patients with carcinoid heart disease usually present with signs of right heart failure caused by tricuspid and pulmonary valve regurgitation; tricuspid valve involvement occurs in 100% of cases, generating moderate to severe regurgitation in 97% of them; the most common findings are thickening of the tricuspid valve leaflets and retraction with shortening of the subvalvular insertions. (2)

Pulmonary valve involvement produces thickening and retraction of the leaflets associated with constriction of the annulus, causing severe regurgitation as-

sociated with mild stenosis. (2)

Mitral and aortic valve involvement is less frequent, with severe regurgitation in approximately 7% of patients diagnosed with carcinoid heart disease. (3) The patient with carcinoid heart disease should be managed by a specialized multidisciplinary team, as this is a systemic disease with multiorgan involvement.

#### Clinical Case N° 1

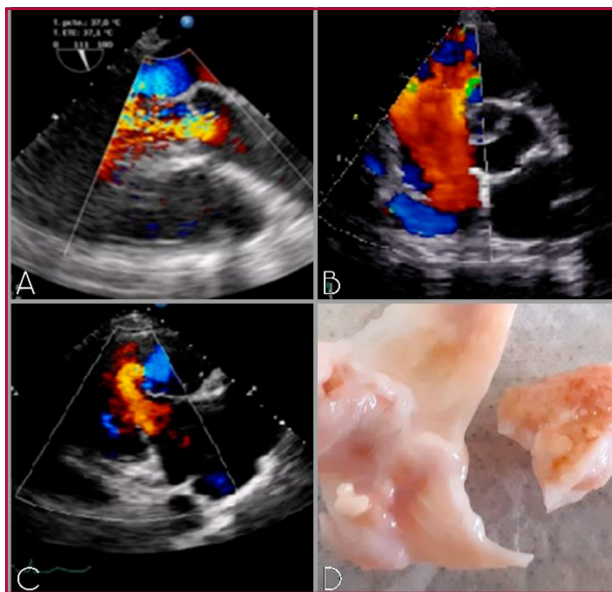
This is a 71-year-old female patient with no cardiovascular risk factors and a history of breast cancer treated at the age of 50, consulting for progressive dyspnea and a diagnosis of small intestine neuroendocrine tumor with multiple liver metastases detected by positron emission tomography (PET). Doppler echocardiography shows severe pulmonary and tricuspid regurgitation, right ventricular enlargement, preserved biventricular function, and left valve involvement with severe aortic regurgitation (Figure 1A, B, C). After a multidisciplinary evaluation, surgical treatment is decided. Intraoperative findings show retraction and fibrous thickening of the valves, typical of carcinoid heart disease (Figure 1D). Tricuspid, pulmonary and aortic valve replacement (N31, N23 and N23, respectively) and aortocoronary bypass grafting with saphenous vein to the right coronary artery due to a lesion diagnosed in the preoperative coronary arteriography are performed. Postoperative course is satisfactory and without complications, and the patient is discharged on the 10th postoperative day. At present, the patient has survived for 3 years, asymptomatic in follow up.

#### Clinical Case N° 2

This is a 50-year-old male patient with a history of hypertension, dyslipidemia and smoking. His oncological history includes pancreatic resection of a carcinoid tumor and radiofrequency ablation of liver metastases, 6 years before the onset of cardiac symptoms (Figure 2A). On this occasion, the patient consults for progressive dyspnea and symptoms of right heart failure. Doppler echocardiography shows severe tricuspid regurgitation associated with moderate pulmonary insufficiency and signs of right ventricular overload, systolic pulmonary pressure of 45mmHg, and preserved tricuspid annular plane systolic excursion (TAPSE) (Figure 2B).

It is decided to perform bioprosthetic tricuspid and pulmonary valve replacement (N31 and N27 respectively) and prophylactic octreotide during surgery (Figure 2C). Postoperative course is satisfactory and without complications, and the patient is discharged on the 6th postoperative day.

Four years after surgery, the patient redevelops progressive dyspnea and is diagnosed with progression of the carcinoid disease and mitral valve involvement. Doppler echocardiography shows mitral valve



**Fig. 1.** A. Free tricuspid regurgitation; B. Severe pulmonary regurgitation; C. Moderate aortic regurgitation. D. Macroscopy of aortic valve, thickened and retracted leaflets.

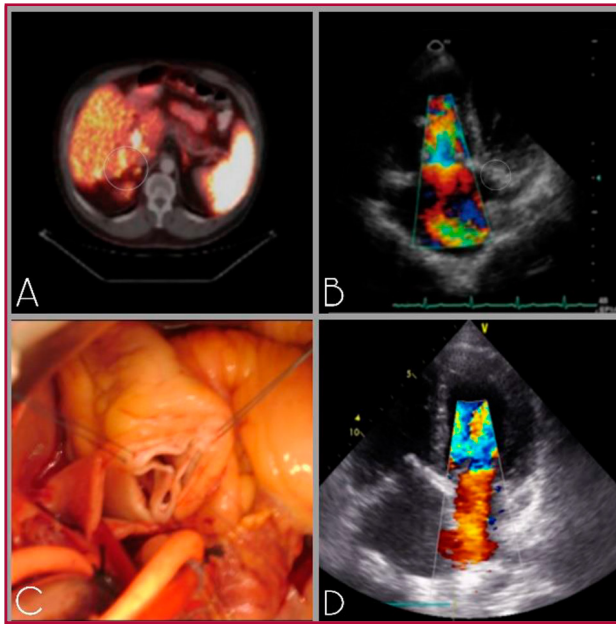
with leaflet retraction and severe mitral regurgitation due to lack of coaptation, with effective regurgitant orifice (ERO) 0.5 (Figure 2D). Tricuspid and pulmonary prostheses are normofunctioning, with adequate gradients. The patient undergoes surgical reintervention, receiving biological mitral valve replacement with an N29 valve; postoperative course is good, and the patient is discharged on the 10th day.

The patient dies two years after the second surgery due to marrow failure associated with oncological disease progression

In patients with severe cardiac valve involvement and controlled systemic disease, valve replacement surgery is the most effective treatment option to reduce morbidity and mortality.

In a retrospective study that included 195 patients undergoing carcinoid heart disease surgery, Connolly et al. demonstrated that the 1-, 5- and 10-year survival rates for valve replacement were 69% (63% to 76%), 35% (28% to 43%) and 24% (18% to 32%), respectively. (4)

In the Nguyen et al. series published by the Mayo Clinic, including 240 patients undergoing operation, early mortality rate was 9% and decreased as time progressed and as operative experience increased. In the present series between 1985 and 1994, early mortality was 29%; however, this risk decreased to 7% between 1995 and 2004, and has been 5% since 2005. Late survival rates at 1, 3 and 5 years, respectively, were 69%, 48% and 34%, with functional class and older age being independent predictors of mortality, suggesting that patients with carcinoid heart disease may benefit from earlier surgical intervention once valve disease



**Fig. 1.** A. PET, diffuse pattern of liver involvement. B. Severe tricuspid regurgitation. C. Intraoperative image of pulmonary valve. D. Severe mitral regurgitation

or right ventricular failure develop symptoms. (5)

Nevertheless, optimal timing of surgery may be challenging to determine in these complex patients whose symptoms occur in combination with gastrointestinal/hepatic impairment. It is often difficult to determine to what degree right ventricular failure is contributing to functional decline and ascites in a patient with carcinoid syndrome and extensive hepatic metastases.

The choice of prosthesis type for valve replacement is still under discussion and should be analyzed individually in each patient; however, considering life expectancy and risk of bleeding associated with oncological disease, bioprostheses would be the first line of choice. (4, 5)

In patients with carcinoid disease, both systemic malignant disease and cardiac involvement should be

addressed simultaneously. The approach should be multidisciplinary at an experienced center, with the participation of cardiologists, hepatologists, cardiovascular surgeons, neuroendocrine tumor specialists, anesthesiologists, and imaging specialists. Patients should be managed on an individual basis, and a holistic approach should be considered.

#### Conflicts of interest

None declared.

(See authors' conflicts of interest forms on the website/ Supplementary material).

#### Ethical considerations

Not applicable

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