

Cardiac Autotransplantation as Treatment Strategy for Malignant Heart Tumors

Primary malignant cardiac tumors, usually sarcomas, represent an infrequent subgroup among cardiac masses. However, they constitute processes with poor prognosis, and surgical treatment is the most favorable therapeutic alternative in terms of survival.

The literature regarding left-sided cardiac sarcomas reveals that patients are subjected to reinterventions for local recurrence, generally related to incomplete resections, probably due to a suboptimal anatomical exposure during surgery, which conditions inadequate resections and technically difficult reconstructions.

Sometimes, to fulfill the objectives of a radical oncological resection and facilitate the reconstruction of the resected cardiac structures, it is necessary to explant the heart in order to resect the tumor with adequate margins and reconstruct the cavities or involved structures, finally reimplanting the heart in bench surgery (cardiac autotransplantation).

This is the case is a female 73-year-old patient, without relevant clinical history who was admitted to hospital due to progressive dyspnea and anemia. In the diagnostic algorithm, the transthoracic echocardiogram showed a dilated left atrium occupied by a 4.8 cm × 2.8 cm immobile, heterogeneous mass, intimately associated with the mitral annulus, which completely filled the left atrial appendage, and severe mitral valve insufficiency with central jet.

A cardiac magnetic resonance performed to complete the mass evaluation revealed the mentioned heterogeneous tumor in weighted T1 and T2 sequences, before and after contrast, as well as in perfusion and late enhancement sequences. No contrast capture was evidenced in the tumor sector protruding to the left atrium, which was interpreted as an added thrombotic component (Figure 1). The same study showed absence of pericardial and pulmonary vein involvement. Prophylactic anticoagulation was started, and a positron-emission computed tomography (PET-CT) of the whole body was performed for local evaluation and search of eventual metastasis.

The PET study showed a hypermetabolic mass of 5.9 cm × 3.4 cm × 2.4 cm (SUV 8.5) in the already known location, focal liver lesions compatible with hemosiderosis and absence of secondaries.

A surgical treatment was decided due to the condition and clinical characteristics of the patient, disease staging and prognosis without resection. Owing to the location of the lesion to resect, in close contact with the mitral annulus, the circumflex artery and the coronary sinus, it was inferred that to perform an adequate oncological resection, the heart should be explanted and reconstructed in bench surgery (ex situ) with subsequent autotransplantation.

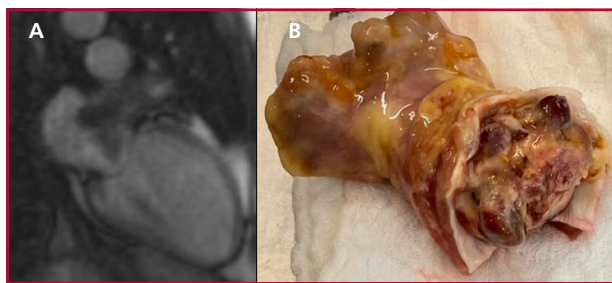


Fig. 1. A. Nuclear magnetic resonance: left atrial sarcoma located in the left atrial appendage. **B.** Surgical specimen with complete lesion resection. The resection margins are evidenced on tissue not affected by the tumor.

Surgery was performed with midline sternotomy and cannulation of both venae cava and aorta

The tumor was explored entering the left atrium by the interatrial sulcus as usually performed for a mitral valve procedure. Absence of pulmonary vein involvement and tumor growth up to the vicinity of the mitral annulus were verified.

Considering that the oncological resection would involve resecting the mitral annulus and part of the mitral valve, and faced with the difficulty to define, through the mentioned approach, the external margin of the resection in relation to the interventricular sulcus structures, it was decided to explant the heart and perform a bench tumor resection.

The venae cava, aorta and pulmonary artery were sectioned and the atriotomy was extended leaving a hood that contained the pulmonary veins. The tumor was resected ex situ (bench surgery), which implied resecting a section of the mitral annulus at the P1 level exposing the atrioventricular sulcus vessels and the ventricular myocardium. The mitral annulus and the left atrium were reconstructed with a bovine pericardial patch and the mitral valve was replaced with a #25 porcine biological prosthesis.

The organ was reimplanted with autotransplantation technique (Figure 2).

Extracorporeal circulation time was 232 min and cross-clamping time 175 min. The postoperative course was in accordance with the magnitude of the procedure, requiring inotropic support for 72 h. Among other events, the patient presented an episode of atrial flutter which was controlled with amiodarone and isolated subfebrile records with negative cultures.

Anatomical pathology reported a grade III undifferentiated pleomorphic sarcoma, which implies a maximum level of malignancy and undifferentiation.

The prevalence of primary cardiac tumors in autopsy series is 0.02%. Among them, 25 % are malignant and 75 % of these are sarcomas. (1) Median survival in published series ranges between 9 and 33 months. (2) Most are clinically silent until a very ad-

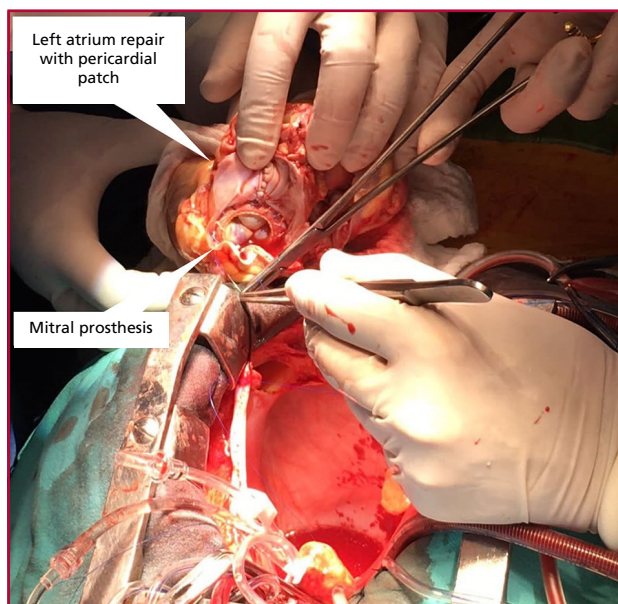


Fig. 2. Cardiac autotransplantation showing the left atrium repaired with bovine pericardium and mitral prosthesis.

vanced stage and are often considered nonresectable due to the proximity to critical structures. However, surgical and imaging techniques have improved allowing more aggressive interventions, which aim to achieve a microscopically negative resection (R0), a situation in which there is clear benefit of survival. (3)

Cardiac autotransplantation is a procedure described many years ago for the resection of tumors with difficult approach or complex intraoperative management. (4)

Along time, the technique was reproduced for the management of this pathology in numerous patients, (5) and the initial results improved in terms of quality of the oncological resection and survival. (6)

With adequate surgical training the technique is reproducible and should be considered a valuable alternative in the therapeutic arsenal to offer opportunities to patients with severe oncological disease and poor prognosis without surgery.

Conflicts of interest

None declared.

(See authors' conflict of interests forms on the web/Additional material.)

Ethical considerations

Not applicable.

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REFERENCES

- Burazor I, Aviel-Ronen S, Imazio M, Markel G, Grossman Y, Yosepovich A, et al. Primary malignancies of the heart and pericardium. *Clin Cardiol* 2014;37:582-8. <https://doi.org/10.1002/clc.22295>
- Chen TW, Loong HH, Srikanthan A. Primary cardiac sarcomas: a multinational retrospective review. *Cancer Med* 2019;8:104-10. <https://doi.org/10.1002/cam4.1897>
- Putnam JB Jr, Sweeney MS, Colon R. Primary cardiac sarcomas. *Ann Thorac Surg*. 1991;51:906-10. [https://doi.org/10.1016/0003-4975\(91\)91003-E](https://doi.org/10.1016/0003-4975(91)91003-E)
- Cooley D, Reardon M, Fraizer O, Angelini P. Human Cardiac Explantation and Autotransplantation: Application in a Patient with a Large Cardiac Pheocromocytoma. *Tex Heart Inst J* 1985;2:171-6.
- Ranlawi B, Al-Jabbari O, Blau L, Davies M, Bruckner B, Blackmon S et al. Autotransplantation for the resection of complex left heart tumors. *Ann Thorac Surg* 2014;98:863-8. <https://doi.org/10.1016/j.athoracsur.2014.04.125>
- Hassan S, Witten J, Collier P, Tong M, Petterson G, Smedira N, et al. Outcomes after resection of primary cardiac sarcoma. *JTCVS Open* 2021;8:384-90. <https://doi.org/10.1016/j.xjon.2021.08.038>

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Compressive Mass in the Anterior Pericardium: Difficulty of the Differential Diagnosis

We report the case of a 79-year-old male patient, smoker, with hypertension, type 2 diabetes, coronary artery bypass surgery in 2009 due to chronic coronary syndrome, and prostatic adenocarcinoma actively followed-up by the Urology Department. The patient was referred to the Emergency Room due to dyspnea on moderate exertion for three weeks and right pleural effusion seen in a chest X-ray. Upon arrival, the patient was hemodynamically stable (blood pressure 153/93 mmHg, heart rate 99 bpm), and oxygen saturation at room air of 89%, with no tachypnea at rest. Physical examination revealed jugular venous distention up to the middle third of the sternocleidomastoid muscle, abolition of the vesicular murmur in the right lung base, and bilateral pitting edema up to both knees.

ECG showed sinus rhythm (89 bpm) with negative T-waves in V1-V4, already present in previous studies. Blood screening showed normal renal function (urea 31 mg/dL, creatinine 0.77 mg/dL, glomerular filtration rate 86 ml/min/1.73 m²) with all ions in range, C-reactive protein 25.98 mg/L, lactate dehydrogenase 950 U/L, creatinine kinase 55 U/L, NT-proBNP 950 pg./mL, ultrasensitive troponin T 25 ng/L, hemoglobin 11.2 g/dL, platelets 335000/mm³, white blood cells 11430/mm³, and D-dimer 4860 ng/mL. To complete the diagnosis, the unsynchronized computed tomography (CT) scan revealed a hypodense lesion over the right cardiac chambers (Figure 1). Neoplasia or hemopericardium due to cardiac rupture or bypass dehiscence were suggested as first possible diagnosis. In view of

these findings, evaluation by the Cardiology Department was requested. Transthoracic echocardiography (TTE) showed a heterogeneous, solid mass in the anterior pericardial sac, with adhesions in the right chambers and compression of the right atrioventricular sulcus, which did not capture echocardiographic contrast (Figure 2). The picture did not suggest a cardiac rupture, not only because of the echocardiographic findings, but also because the patient did not experience chest pain and was hemodynamically stable, and ECG did not show abnormalities suggestive of acute ischemia, making bypass dehiscence unlikely.

Given the discrepancies, a synchronized CT scan was performed for better characterization of the lesion, which showed an 8.5 x 10 cm right precardiac mass, apparently depending on the pericardium, and showing enhancement after intravenous contrast

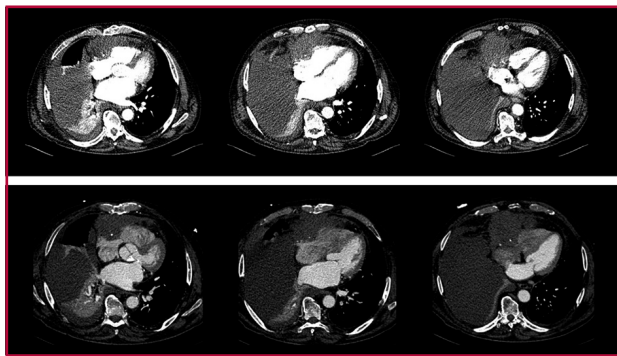


Fig. 1. CT scan. *Top:* Unsynchronized CT scan, with a hypodense lesion over the right cardiac chambers and severe right pleural effusion with associated collapse. *Bottom:* Synchronized CT scan, with a right precardiac mass of 8.5 x 10 cm, enhanced after contrast injection. The tumor exerts mass effect and possibly infiltrates the right atrium, in contact with anterior chest wall and possible infiltration of the right ventricle.

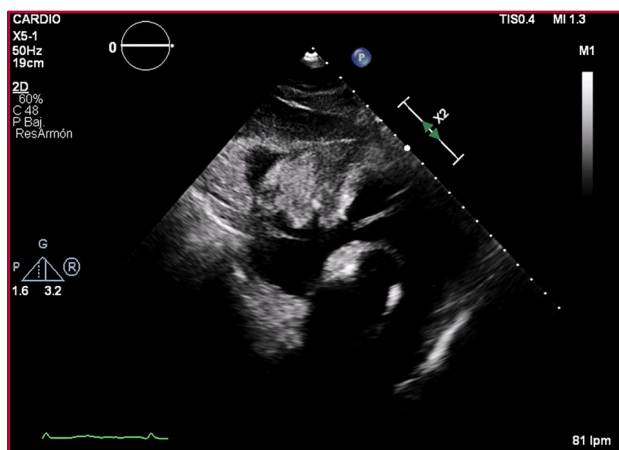


Fig. 1. Transthoracic echocardiography, subcostal access. Heterogeneous, solid mass in the anterior pericardial sac, compressing the atrioventricular groove and appearing to adhere to the ventricular wall.

(Figure 1). These findings raised the differential diagnosis of metastasis or primary pericardial neoplasia.

The differential diagnosis of mediastinal masses is primarily based on the location of the mass, its composition and the age of the patient. (1) Different radiological techniques, including CT scan and cardiac magnetic resonance (CMR), are of significant diagnostic value. Considering that the lesion was located in the anterior wall of the heart, in contact with the anterior chest wall, a broad differential diagnosis including the different lesions at the level of the anterior mediastinum and the tissue-dependent masses in the pericardium was proposed.

Unlike in our patient, thymomas appear as an oval, homogeneous mass, with well-defined contours in CT scan. Calcifications and cystic areas are usually present in thyroid goiters and teratomas, (1-2) and considering that CT findings showed no calcifications in our patient, these two entities seemed unlikely. Lymphomas account for 20% of mediastinal tumors in adults, and Hodgkin's lymphomas are the most common subtype. (1-3) Within this subtype, mediastinal large B-cell lymphoma constitutes an independent entity within the classification of malignant lymphoid neoplasms, with a frequency estimated at 2-3% of non-Hodgkin's lymphomas and between 6-10% of large B-cell lymphomas. This tumor usually occurs as a rapidly expanding mediastinal mass and may be associated with pleural or pericardial effusion. (4, 5)

The patient was admitted to the Cardiology Department to complete assessment. An ultrasound-guided thoracentesis of the pleural effusion was performed, and a serosanguineous fluid consistent with an exudate (Light's criteria) was obtained, showing a hypercellular area in the cytological examination, with characteristics indicative of a B lymphoproliferative process. To complete the study, a core biopsy of the mediastinal mass was performed, confirming the diagnosis of primary mediastinal large B-cell lymphoma. Finally, the first cycle of chemotherapy was started with rituximab, cyclophosphamide, non-pegylated liposomal doxorubicin, vincristine, and prednisolone. He is still on treatment.

Conflicts of interest

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REFERENCES

1. Prosch H, Röhrich S, Tekin ZN, Ebner L. The role of radiological imaging for masses in the prevascular mediastinum in clinical practice. *J Thorac Dis* 2020;12:7591-7. <https://doi.org/10.21037/jtd-20-964>
2. Nakazono T, Yamaguchi K, Egashira R, Mizuguchi M, Irie H. Anterior mediastinal lesions: CT and MRI features and differential diagnosis. *Jpn J Radiol* 2021;39:101-17. <https://doi.org/10.1007/s11604-020-01031-2>
3. Pfau D, Smith DA, Beck R, Gilani KA, Gupta A, Caimi P. Primary Mediastinal Large B-Cell Lymphoma: A Review for Radiologists. *AJR Am J Roentgenol* 2019;213:W194-W210. <https://doi.org/10.2214/AJR.19.21225>
4. Lees C, Keane C, Gandhi MK, Gunawardana J. Biology and therapy of primary mediastinal B-cell lymphoma: current status and future directions. *Br J Haematol* 2019;185:25-41. <https://doi.org/10.1111/bjh.15778>
5. Martelli M, Ferreri A, Di Rocco A, Ansuinelli M, Johnson PW. Primary mediastinal large B-cell lymphoma. *Crit Rev Oncol Hematol* 2017;113:318-27. <https://doi.org/10.1016/j.critrevonc.2017.01.009>

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Native Tricuspid Valve Infective Endocarditis

Right-sided infective endocarditis is a rare but potentially fatal disease. It comprises 5-10% of the total number of infective endocarditis events. It is most frequently associated with intravenous drug use, and occurs less frequently in patients with venous access, intravascular devices or underlying congenital heart disease, and exceptionally in non-addict patients or in patients without cardiac malformations. (1)

We report the case of a 58-year-old male patient with hypertension, dyslipidemia, and a history of aortic valve replacement with mechanical prosthesis due to aortic stenosis in 2019.

He is admitted to the general ward for febrile syndrome under study. Cardiac physical examination reveals neither changes in heart sounds nor signs of congestive heart failure. ECG shows sinus tachycardia and first-degree atrioventricular block (PR interval 270 msec).

Blood screening on admission shows white blood

cells 31 840/mm³ (neutrophils 96%, lymphocytes 1.7%), C-reactive protein 58.9 mg/L (normal range 0-5), procalcitonin 4.55 ng/mL (normal range 0-0.1), total bilirubin 1.33 mg/dL, indirect bilirubin 0.83 mg/dL, and direct bilirubin 0.50 mg/dL.

During hospitalization, methicillin-susceptible *Staphylococcus aureus* is detected in blood cultures. Due to suspicion of infective endocarditis, a transesophageal echocardiogram is performed, that detects no vegetations, and normal mechanical valve function. On the fourth day of antibiotics, further blood cultures detect no bacterial growth; on the tenth day, transesophageal echocardiography shows no vegetations in the heart valves, ruling out infective endocarditis. After receiving intravenous antibiotics during 14 days, patient is discharged.

A week later, he is readmitted for fever and general malaise; ECG reveals further PR interval prolongation (270 msec) (Figure 1A). Blood screening shows white blood cells 11 870/mm³ (neutrophils 87%, lymphocytes 4.7%), C-reactive protein 25.6 mg/L, procalcitonin 0.19 ng/mL, erythrocyte sedimentation rate 32 mm/h. Methicillin-susceptible *Staphylococcus aureus* is isolated in follow-up blood cultures. Transthoracic echocardiography reveals a 0.6 cm x 0.6 cm mobile image at the tricuspid valve level. Transesophageal echocardiography (TEE) confirms 0.9 x 0.6 cm vegetation at the level of the septal leaflet, mild tricuspid regurgitation, and normal prosthetic valve function (Figure 2).

A conservative approach with intravenous cefazolin for 6 weeks is followed. First-degree AV block improves by the fifth week of antibiotic treatment (Figure 1B). Follow-up transthoracic echocardiograms on the second and sixth weeks of treatment show no evidence of tricuspid vegetation.

Outpatient positron-emission tomography/computed tomography (PET/CT) for suspected prosthetic valve involvement reveals moderate diffuse radiotracer uptake at the level of the replaced aortic valve, suggesting the absence of an active infectious process, given the absence of a dominant focus with increased concentration of the contrast material and SUVmax 3.5 (Figure 3).

Right-sided infective endocarditis is common in injecting drug addicts and in patients with cardiac malformations; it is a potentially serious condition, with a mortality rate between 23 and 31%. Simultaneous left and right-sided endocarditis comprises 13% of the cases, whereas right-sided endocarditis alone affects 10%. (1, 2)

Isolated native tricuspid valve endocarditis (NTVE) usually occurs spontaneously, without evident history of dental or surgical procedures; however, the skin is usually the most common portal of entry (particularly in the case of *S. Aureus*). In this clinical case, the predisposing factor could not be identified. *Staphylococcus aureus* is the most commonly isolated infectious agent (70% of cases), followed by *Streptococcus* and *Enterococcus*. (3)

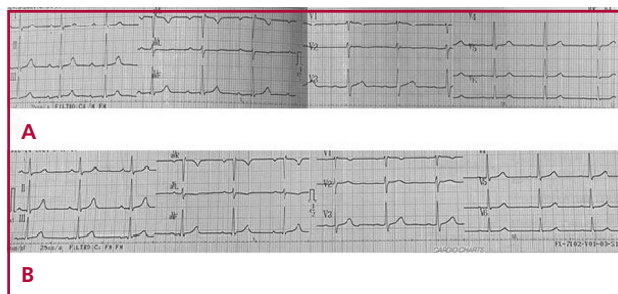


Fig. 1. A. ECG: Prolonged PR interval (270 msec). B. ECG: Normal PR interval (200 msec).

The clinical presentation invariably consists of persistent fever associated to pulmonary events, anemia and microscopic hematuria (tricuspid syndrome of Nandakumar and Raju). The absence of peripheral stigmata of endocarditis or relevant murmurs in most cases is noteworthy. (4)

If fever is persistent (if it remains after a 2-week course of antibiotics) it is usually associated with perivalvular extension of infection, new septic emboli or superimposed nosocomial infection. The clinical picture, positive findings on blood culture and echocardiography are the main diagnostic tools in NTVE. (4)

The usefulness of PET/CT is significantly greater for prosthetic valve endocarditis than for native valve infective endocarditis and is an excellent alternative in case of negative or doubtful ultrasound scans. Integrating PET/CT as a diagnostic tool in endocarditis allows for reclassification of 76% of patients with

prosthetic-valve infective endocarditis from "possible" to "definite". (5)

Eighty percent of isolated NTVE patients are successfully treated with medical therapy. However, surgery is recommended in uncontrolled infection or right heart failure with tricuspid regurgitation refractory to treatment. Surgical treatment repairs the valve dysfunction and eliminates the infectious focus, thus contributing to reduce mortality associated with heart failure. (6)

Regarding prognosis, a high success rate is achieved with medical treatment (antibiotics), the development of heart failure is uncommon, and only 25% of cases require valve replacement or surgery. (1) Mortality associated with isolated NTVE is lower than that reported for endocarditis with a predisposing condition. (6)

This case suggests the need to consider isolated NTVE, its clinical presentation, treatment, and prognosis, as well as the usefulness of PET/CT to confirm prosthetic valve involvement.



Fig. 2. TEE: View at 60°. Tricuspid septal leaflet vegetation is observed.



Fig. 3. PET/CT with fluorine-18 deoxyglucose: transverse projection. Diffuse uptake in the aortic prosthesis is observed.

Conflicts of interest

None declared.

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Ethical considerations

Not applicable.

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REFERENCES

1. Zaldívar AA, Cardoso AA, Ramon RD. Endocarditis Infecciosa Derecha. Presentación de un caso. *Rev Cub Cardiol Cirug Cardiovasc* 2019;25(4).
2. Pérez Domínguez JA, Aguilar Almaguer O, González Céspedes JC, Escandell Reyes A, Leyva Castro R, Rodríguez Peña MM. Complicaciones sistémicas en endocarditis infecciosa de válvula tricúspide. *Multimed* 2019;23:543-51.
3. Salamanca MA. Endocarditis tricuspídea secundaria a infección asociada a catéter venoso central. Reporte de dos casos. In *Anales de la Facultad de Medicina* 2020;81:330-32. UNMSM. Facultad de Medicina.
4. Alkan G, Emiroglu M, Sert A, Kartal A, Öc M. Endocarditis infecciosa de la válvula tricúspide asociada con meningitis aséptica: presentación infrecuente en una niña. *Arch Argent Pediatr* 2020:e22-e25.
5. Ladrón-de-Guevara H, Canelo L, Bitar H, Ramón Soto J. Imágenes en endocarditis infecciosa: No todo es ecocardiografía. *Rev Chil infectol*, 2021;38:260-70.
6. Álvarez F, Torrez J, Galleguillos G, Saavedra J. Endocarditis infecciosa cámaras derechas. Reporte de un caso. *Rev Chil Anest* 2021;50. *Rev Argent Cardiol* 2023;91:94-96. <http://dx.doi.org/10.7775/rac.v91.i1.20604>