

A Rare Case of Simultaneous Both-side Heart Valvular Infective Endocarditis on Ventricular Septal Defect

ABSTRACT

IE is a rare pathology with an annual incidence of approximately 10 cases/100,000 inhabitants and in spite of staphylococci pathogens, streptococci are the most involved pathogens, acute articular rheumatism remains the most frequent etiology in developing countries. It can attack a healthy heart, but most often seen in a diseased heart. The incidence of bilateral right and left heart IE is significantly lower, accounting for 56% to 10% of all IE cases. Only few cases of bilateral IE on the native valve have been found in the literature. Echocardiography, whether transthoracic (TTE) or transesophageal (TOE), is a crucial examination in the management and follow-up of any IE. Vegetations are the specific lesions of endocarditis. The authors report a case of a multiple native heart valvular infective endocarditis simultaneously attacking the left and right-sides heart valves confirmed by transthoracic echocardiography and cultures in a patient poorly followed-up for non-cyanotic congenital heart disease without hemodynamic impact, diagnosed with chronic kidney disease undergoing hemodialysis on a jugular catheter. A coupled medical and surgical treatment was conducted in the patient with a very satisfying clinical outcome and less complications after an open-heart intervention.

Keywords: Infective endocarditis; ventricular septal defect; cardiology; infectious attack.

ABBREVIATIONS

AV-Bloc	: Atrio-ventricular Bloc
Bpm	: Beats per Minute
BP	: Blood Pressure
CKD	: Chronic Kidney Disease
CoNS	: Coagulase Negative Staphylococcus
ECG	: Electrocardiogram
ESRD	: End Stage Renal Disease
HBV/HBC	: Hepatitis B/ Hepatitis C
HIV	: Human Immunodeficiency Virus
IE	: Infectious Endocarditis
LVH	: Left Ventricle Hypertrophy
NVE	: Native Valve Endocarditis
PHT	: Pressure Half Time
RF	: Rheumatoid Factor
RIE	: Right Side Infective Endocarditis
TTE	: Transthoracic Echocardiography
TOE	: Transesophageal Echocardiography
VSD	: Ventricular Septal Defect

1. INTRODUCTION

Infectious endocarditis (IE) is a microbial infection of one or more heart valves and its endothelial lining by a germ. It is a rare pathology with an annual incidence of approximately 10

cases/100,000 inhabitants [1] and in spite of staphylococci pathogens, streptococci are the most involved pathogens, acute articular rheumatism remains the most frequent etiology in developing countries. It can attack a healthy heart, but most often seen in a diseased heart [2]; and although IE on rheumatic heart valves has always occupied the first place in developing countries [3]. Despite therapeutic progress and in particular the improvement of surgical techniques, endocarditis remains a serious disease with a high mortality rate of 30% [4]. However, its epidemiology has recently changed with the increase in Staphylococcal pathogen infections.

IE most frequently involves left-sided cardiac structures. Simultaneous left and right-sided native valve IE is uncommon. *Staphylococcus aureus* (*S. aureus*) is the predominant organism for both-sided IE. It occurs predominantly in patients with valvular heart disease, a prosthetic valve, intravenous drug abuse, cardiac device insertion, or congenital heart disease. Shunt diseases are common risk factors of both-sided IE. *Streptococcus anginosus* is usually associated with pyogenic infections, but remains a rare cause of IE [5].

We report an extremely rare case of a multiple native heart valvular infective endocarditis in a

patient poorly followed-up for non-cyanotic congenital heart disease diagnosed with chronic kidney disease (CKD) undergoing hemodialysis on a jugular catheter which was not previously reported in the literature.

2. CASE REPORT

We report the case of a 35-year-old young man from Guinea admitted for NYHA stage III dyspnea accompanied by palpitation, atypical chest pain and especially abdominal distension.

The patient has a history of a restrictive ventricular septal defect (VSD) measuring 4 mm wide for 4 years without hemodynamic impact and end-stage renal disease (ESRD) undergoing hemodialysis at the rate of 2 sessions per week.

Somatic examination finds a stable patient with a BP of 120 mmHg systolic pressure and 80 mmHg diastolic pressure, mild tachycardia at 112 bpm, body temperature of 38.5°C, saturated at 95% in the open air. The cardiovascular examination found signs of congestive heart failure with the predominance of right heart failure signs such as abdominal distension evoking ascites, jugular turgor and lower limbs edema going up to the thighs. Cardiac auscultation found a multifocal systolic-diastolic murmur and the rest of the examination in search

of the portal pathogen entry was unremarkable except for a degraded oral dentition state. The ECG had shown a regular sinus rhythm associated with a first-degree AV-Bloc with left ventricular hypertrophy (LVH) (Fig. 1).

Chest X-ray reveals cardiomegaly without lung parenchymal abnormalities (Fig. 2).

Transthoracic echocardiography revealed a dilated, hypertrophied left ventricle with conserved global and segmental contractility with 59% of LVEF by Simpson biplane, bi-atrial dilation, restrictive infundibular VSD (Fig. 3). Thickened mitral valve, remodeled with perforation of the large mitral valve associated with an echogenic formation responsible for a severe mitral leak (Fig. 4), aortic valve of which seated vegetations at the level all the 3 cusps of the valve with false aneurysm of the right anterior cusp responsible for a severe aortic leak (PHT=58ms) (Fig. 5), dilacerated pulmonary valve seated with mobile echogenic formations, measuring 27 mm long axis responsible for a severe pulmonary leak (PHT=58 mm) and moderate tricuspid leak estimating the systolic pulmonary pressure at 44 mmHg with seated vegetation on the anterior leaflet of the tricuspid valve (Fig. 6). The RV was dilated with a longitudinal systolic dysfunction without pericardial effusion.

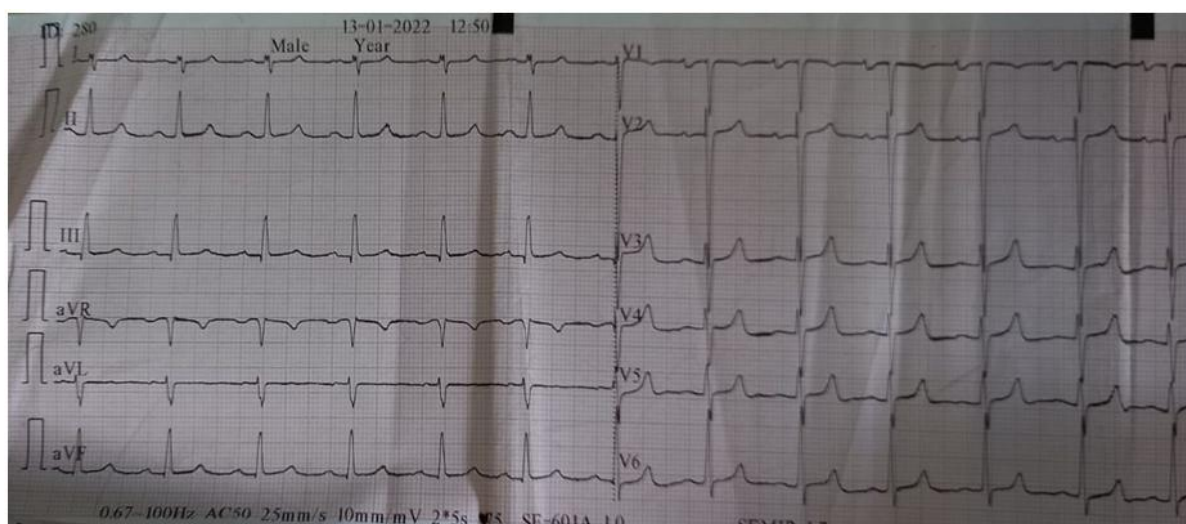


Fig. 1. Electrocardiogram (ECG): Regular sinus rhythm, 1st degree AV-Bloc associated with electrical left ventricular hypertrophy (LVH)

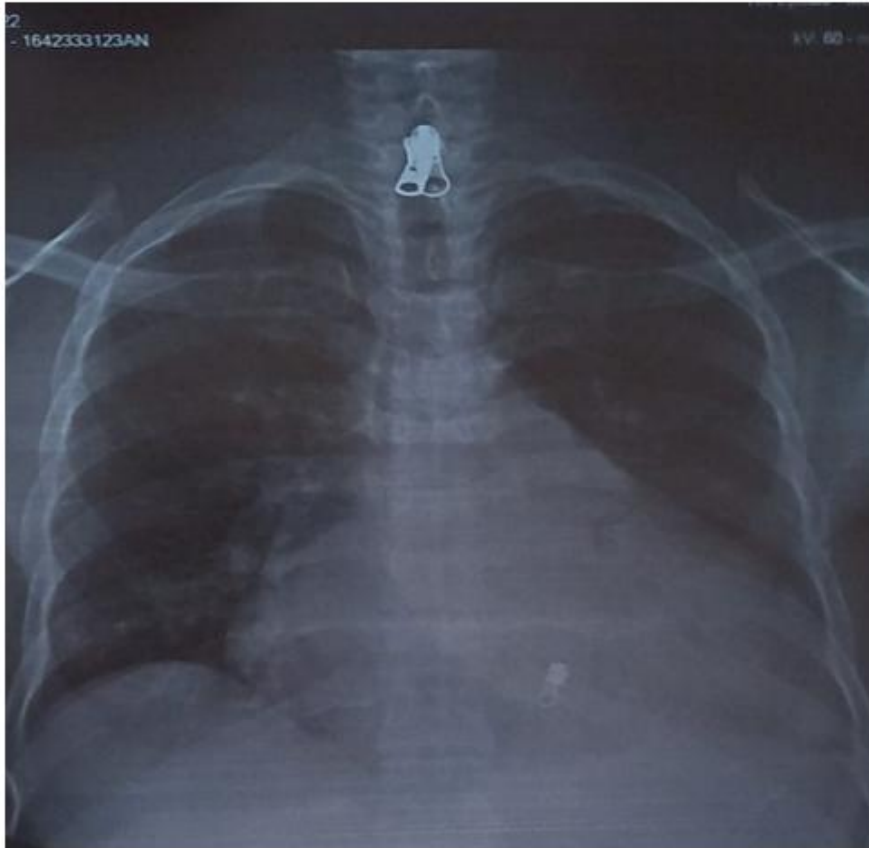


Fig. 2. Frontal chest X-ray: Cardiomegaly with ventricular hypertrophy associated with obliteration of the bottom of the left costo-diaphragmatic sac

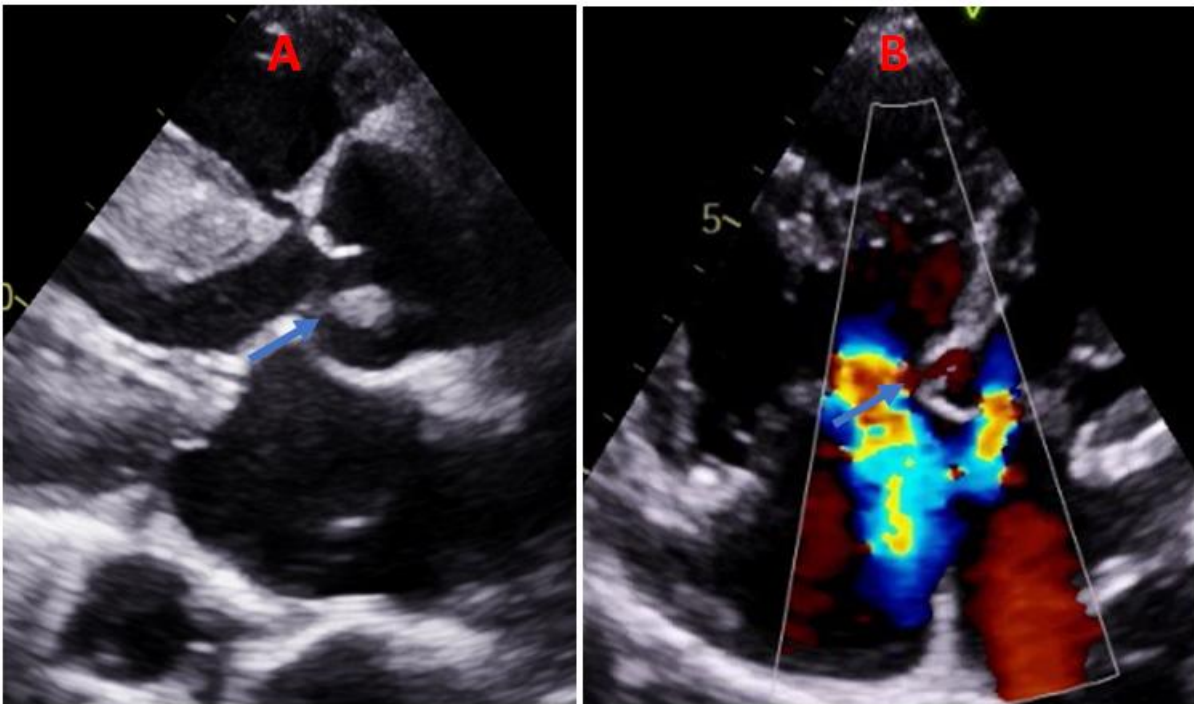


Fig. 3. TTE: A- Long-axis parasternal view: shows a restrictive infundibular VSD with a left-to-right shunt of measuring 4mm and echogenic formation on the posterior cusps of the aortic

valve. B- Apical 4 chambers view: showing the left-right shunt of the infundibular VSD with color Doppler

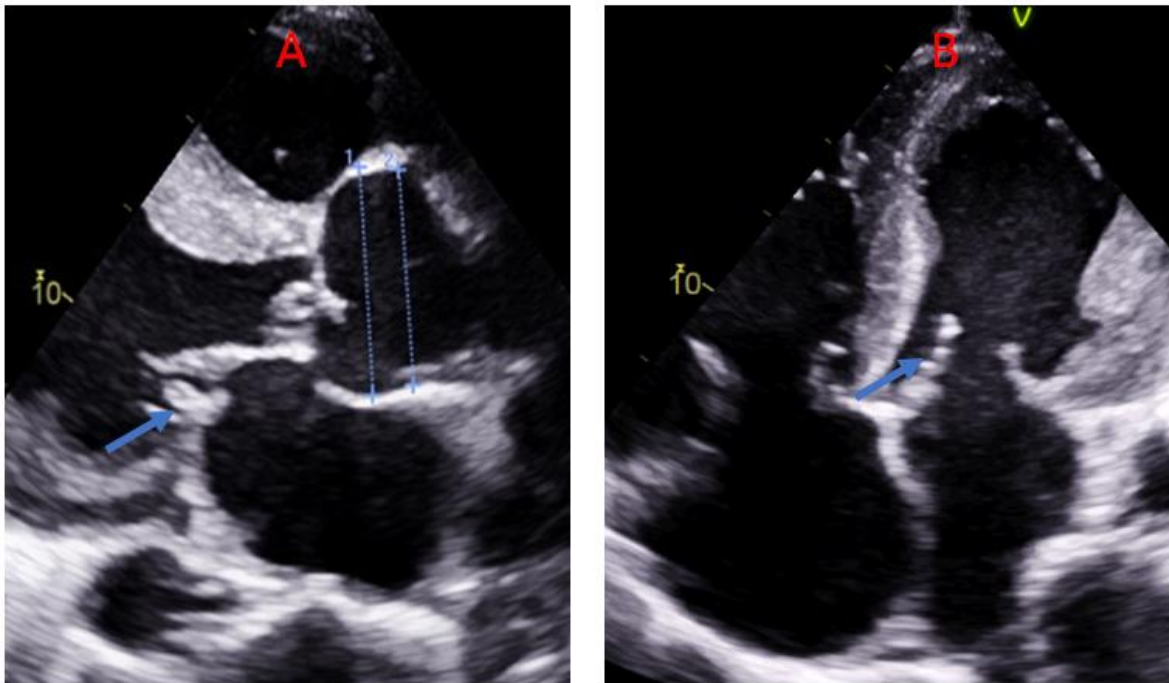


Fig. 4. TTE: Long-axis parasternal view: A- thickened mitral valve, remodeled, site of an echogenic formation (vegetation) mobile located on the small mitral valve
B-TTE: Apical 4 chambers view: showing perforation of the large mitral valve and dilation of the right chambers

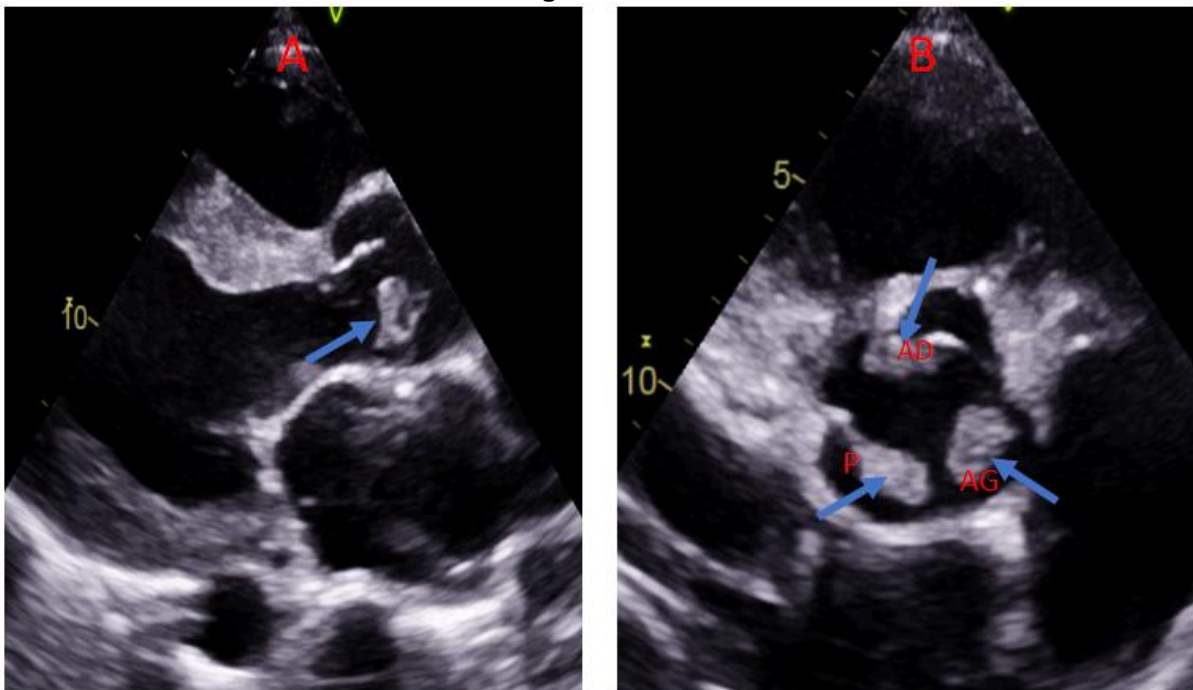
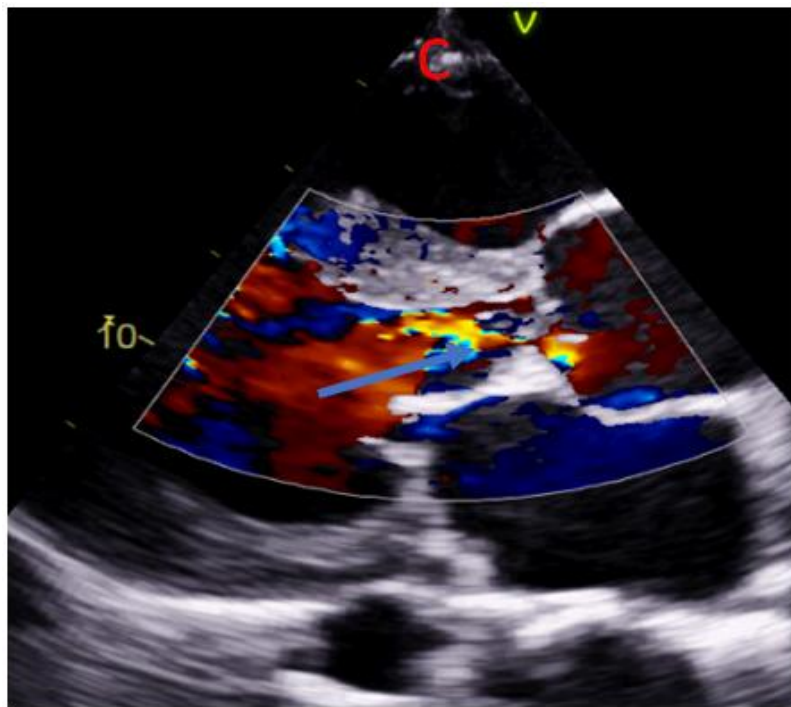


Fig. 5. Transthoracic echocardiography (TTE): A- long-axis parasternal view: Hypertrophy of the LV, echogenic image visible in systolic time on the posterior cusps

B- Parasternal short axis view: echogenic image on the antero-right (A-R), antero-left and posterior cups with a pseudoaneurysm of the A-R cusp



C- Transthoracic echocardiography: Parasternal short-axis view: color Doppler, flow of severe aortic leak

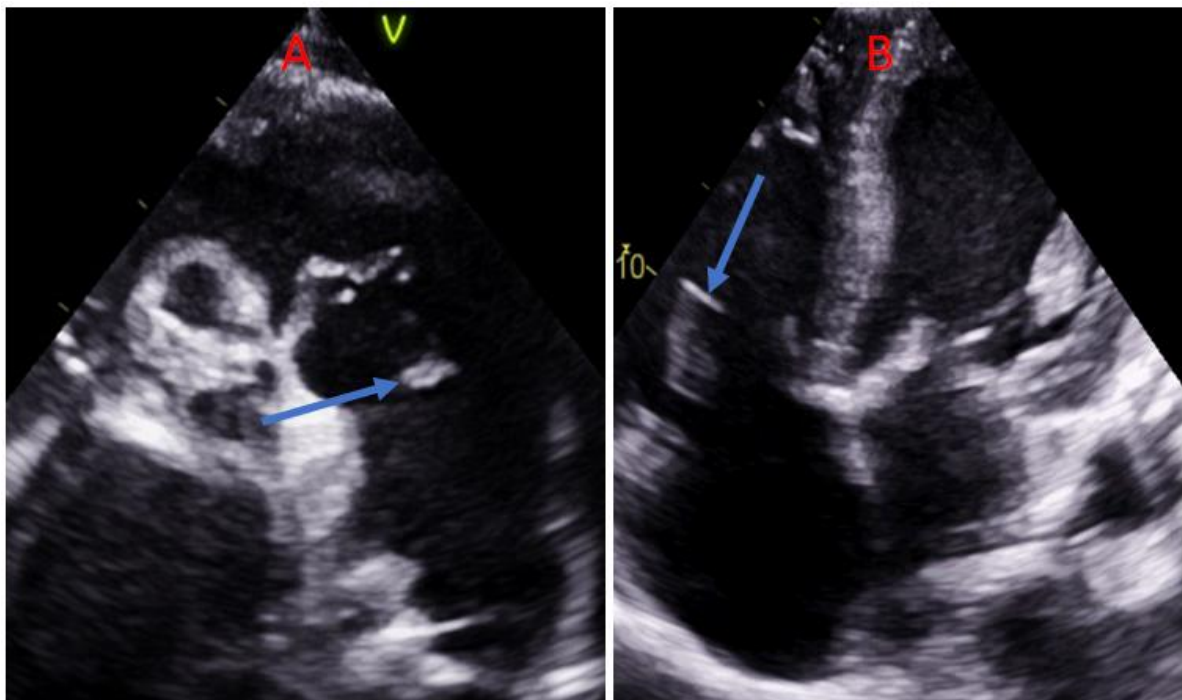


Fig. 6. Transthoracic echocardiography: A- parasternal short axis view: dilacerated pulmonary valve seated with mobile echogenic formations, measuring 27 mm long axis responsible for severe pulmonary leakage.

B- Apical 4 chambers view: shows an echogenic formation on the anterior leaflet of the tricuspid valve

The biological assessment concluded a microcytic hypochromic anemia at 6.7g / dl, altered infectious assessment with hyperleukocytosis at 11000 elements with predominant neutrophilia cells, C-reactive protein raised at 118 mg / l and positive procalcitonin at 0.6 ng / l.

In view of the clinical, ultrasound and biological findings, infective endocarditis with multiple heart valve attack was suspected, the blood cultures carried out came positive with the presence of a gram-positive cocci, a coagulase-type negative staphylococcus (CoNS) multidrug resistance to antibiotics. The search for germs on the dialysis device at the right jugular level after ablation is positive for the same germ.

The endocarditis extension assessment is completed by an immunology assessment (RF, Ac Anti DNA, etc.) and viral serologies (HBV,

HCV, HIV, TPHA/VDRL) which turned out to be negative.

The patient was put on bi-antibiotic therapy based on vancomycin 100mg after each dialysis session and Imipenem 2g in per-dialysis session associated with fluconazole 400 mg. The surgical indication was posed in the patient for a double mitro-aortic valve replacement and a valvuloplasty of the pulmonary valve.

A good clinical evolution was marked in the patient after 30 days of bi-antibiotic well adapted therapy. The patient was operated by double replacement of the mitro-aortic valve by mechanical prosthesis and valvuloplasty of the pulmonary valve (Fig. 7). The postoperative follow-up was without complications and the patient put on an oral anticoagulant treatment based on AVK (sintrom) with an INR objective between 2.5-3.5.

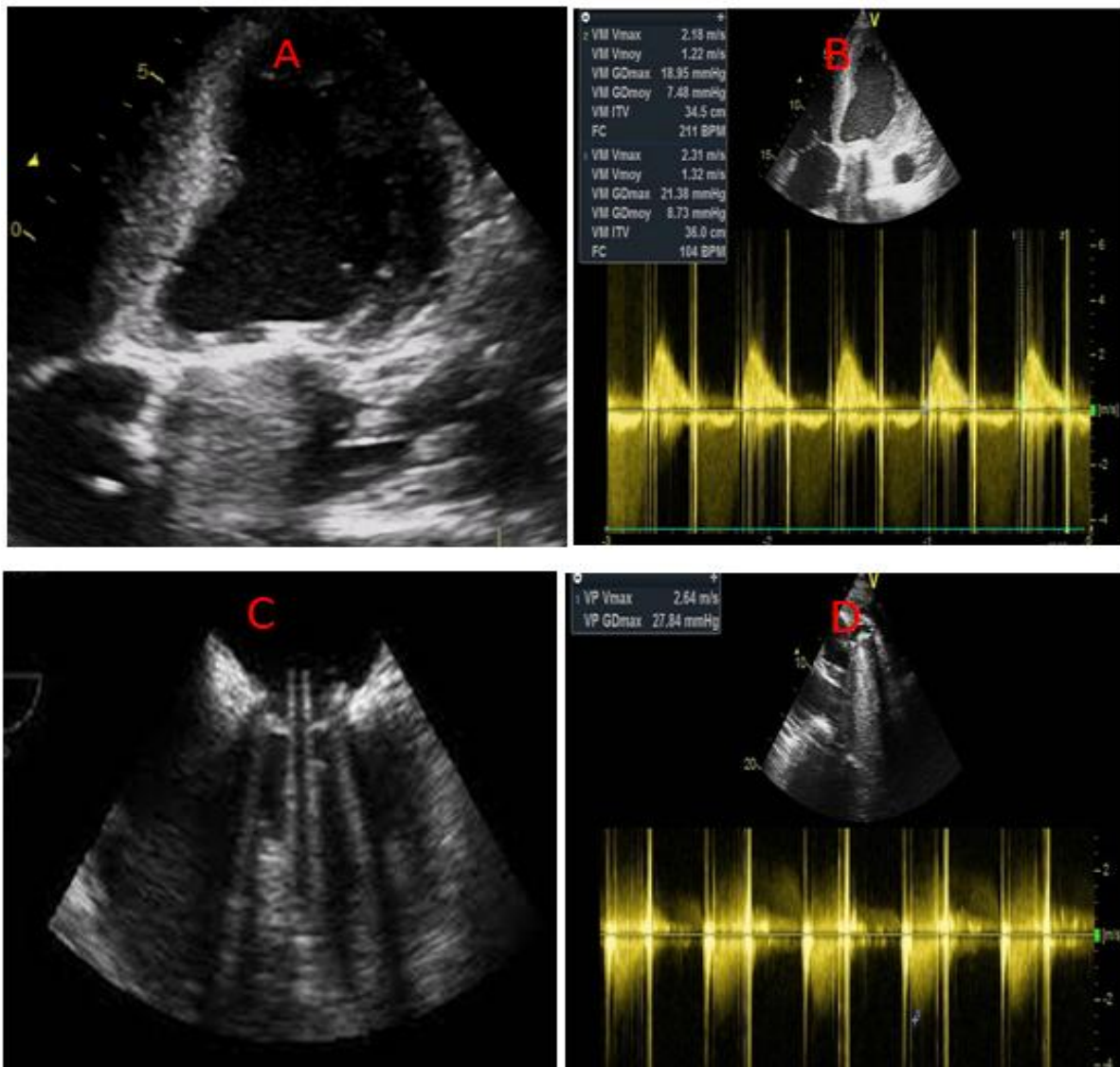


Fig. 7. A-TTE: mechanical prosthesis in mitral position with fin artifact visualized in the LA chamber. b-TTE: Mean gradient of the mitral prosthesis 24hrs after valve replacement. c-TOE: Showing mechanical prosthesis in aortic position with visualization of the prosthetic fins. d-TTE: Mean gradient of pulmonary valve plasty

3. DISCUSSION

We present here a rare case of IE that has not been reported in the literature before. This case demonstrates infective endocarditis affecting all left and right heart side native valves confirmed by transthoracic echocardiography and blood cultures.

The incidence of bilateral right and left heart IE is significantly lower, accounting for 56% to 10% of all IE cases [6]. Only few cases of bilateral IE on the native valve have been found in the literature [7,8].

Infectious endocarditis (IE) is an infectious attack of the endocardium causing mainly valvular damage, responsible for mortality and significant morbidity. It can occur on a healthy or pathological heart. Among the heart diseases at risk, congenital heart disease of which VSD is the most frequent predisposing factor [9]. Various studies and registries have also found that among congenital heart disease causing IE, VSD comes first; patients who are carriers are six times more likely to have an IE than the general population [10,11,12]. In our case the only specificity is the infundibular location of the VSD.

S. aureus was the most common pathogen in bilateral IE [6]. In our case, the isolated pathogen was a multidrug-resistant coagulase negative staphylococcus. IE occurs predominantly in patients with valvular heart disease, a prosthetic valve, intravenous drug abuse, medical device implantation, or congenital heart disease. RIE occurs predominantly in intravenous drug users [13] Intracardiac or extracardiac shunt diseases (e.g., ventricular septal defects, patent ductus arteriosus) are common risk factors of both-sided IE [14]. The shunting can result in the development of IE from single side to both sides of the heart involvement. The pathogenesis of IE in our patient was related to the underlying congenital heart disease (VSD) and the percutaneous implantable hemodialysis device.

Echocardiography, whether transthoracic (TTE) or transesophageal (TOE), is a crucial examination in the management and follow-up of any IE. Vegetations are the specific lesions of endocarditis. It is important to identify early development of vegetations for optimal treatment. In the meantime, the examiner should beware of pitfalls in echocardiography, otherwise an erroneous diagnosis could be made. Vegetations must be distinguished from thrombi, cardiac tumour, myxoma, trophus as well as growths of Lambli [6]. In our case the vegetations are more manifested with their mobile character except that on the tricuspid valve which was mistaken for a thrombi.

Early and accurate diagnosis of IE is crucial to achieve the best therapeutic strategy. Surgical intervention is superior to medical intervention for right heart valve involvement because interventional surgery can adequately remove all infected tissue [15]. Although antibiotic therapy is often effective at treating NVE, surgical intervention is necessary in 20% to 40% of patient [16,17]. In patients with endocarditis on the left side of the heart who are in stable condition, changing to oral antibiotic treatment was noninferior to continued intravenous antibiotic treatment [18] whilst in RIE, surgical intervention is superior to medical intervention [5]. In our case, with the involvement of all the heart valves, a medical treatment based on dual antibiotics therapy by intravenously was conducted for a 30 days period with a good clinical outcome observed before heart valve surgery replacement.

VSDs are congenital heart defects that increase the risk of IE in the adult population, but there is very little discussion of this anomaly as a

complication of IE reported in the literature [5,19]. In our case, the patient presents a higher risk of IE in front of congenital heart disease and the presence of a percutaneous hemodialysis device.

VSD is a benign cardiac lesion, the prognosis of which can be severely compromised by infectious endocarditis: surgical repair reduces the risk but does not totally exclude it because of minor associated abnormalities [20]. It is reported that surgical intervention has a better prognosis in the case of IE on congenital heart disease type VSD than medical treatment alone [16]. In our case, the combination of medical and surgical treatment was of good clinical outcome with desperation of the signs of congestive heart failure.

Prophylactic antibiotic therapy was indicated to our patient before certain surgical procedure, particularly dental intervention as his VSD was not closed during the IE surgery because of its small diameter.

4. CONCLUSION

Simultaneous left and right heart infective endocarditis is a rare pathology, especially when all heart valves are affected. VSD, intra or extracardiac devices can be the triggering risk factors, hence the interest of suspecting the diagnosis in subjects with acute or prolonged fever.

Treatment with probabilistic and or specific antibiotics for the identified germ must be as early as possible. The coupled medical-surgical treatment showed a better prognosis in the bivalvular attack which was also noted in our own case with a good clinical outcome.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

ACKNOWLEDGEMENT

I thank the whole Department of Cardiology P37, university teaching hospital Ibn Rochd Casablanca for their outstanding collaboration in the support of this work.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Fabio Chirillo, Pompilio Faggiano, Moreno Cecconi, Antonella, Moreo, Angelo Squeri, Oscar Gaddi, Enrico Cecchi. Predisposing cardiac conditions, interventional procedures, and antibiotic prophylaxis among patients with infective endocarditis. *AmHeart J.* 2016;179:42-50.
2. Gilbert Habib, Patrizio Lancellotti, Manuel J Antunes, Maria Grazia Bongiorno, Jean-Paul Casalta, Francesco Del Zotti, Raluca Dulgheru, Gebrine El Khoury, Paola Anna Erba, Bernard Lung, Jose M. Miro, Barbara J Mulder, Edyta Plonska Gosciniak, Susanna Price, Jolien Roos-Hesselink, Ulrika Snygg Martin, Franck Thuny, Pilar Tornos Mas, Isidre Vilacosta, Jose Luis Zamorano. 2015 ESC Guidelines for the management of infective endocarditis. *Rev Esp Cardiol.* 2016;69:69.
3. Maharaj Breminand, Parrish Andrew. Prevention of infective endocarditis in developing countries. *Cardiovasc J Afr.* 2012;23:303-5.
4. Lucie Duarte, Adrien Bouglé. Infective endocarditis. *J. Anrea.* 2021;7:396-409.
5. Jian-Hong Pan, MD. Rare simultaneous left and right-sided native valve infective Endocarditis Caused by Rare Bacterium *Int Heart J.* 2019;18:347.
6. Xie J, Liu S, Yang J, Xu J, Zhu G. Inaccuracy of transthoracic echocardiography for the identification of right-sided vegetation in patients with no history of intravenous drug abuse or cardiac device insertion. *J Int Med Res.* 2014;42:837-48.
7. Oylumlu M, Ercan S, Basanalan F, Davutoglu V. Both-sided native valve endocarditis in an intravenous drug misuser. *BMJ Case Rep.* 2013; bcr2013201980.
8. Tyagi S, Patki S, Vaideeswar P, Meshram V. Both-sided native valve infective endocarditis in a drug addict with incidental pneumoconiosis. *J Forensic Leg Med.* 2018;58:41-3.
9. Hanane Boussir, Amine Ghalem, Nabila Ismaili, Noha El Ouafi. Infective endocarditis in patients with ventricular septal defect: The role of echocardiography and prophylactic antibiotic therapy. *Pan African Med J.* 2016;25:154
10. Baltimore RS, Gewitz M, Baddour LM, Beerman LB, Jackson MA, Lockhart PB, et al. Infective endocarditis in childhood: 2015 update: A Scientific statement from the American Heart Association. *Circulation.* 2015;132:1487-515.
11. Tribak M, Konaté M, Elhassani A, Mahfoudi L, Jaabari I, Elkenassi F, et al. Aortic infective endocarditis: Value of surgery. About 48 cases. *Ann Cardiol Angeiol.* 2016;65:15-20.
12. Berglund E, Johansson B, Dellborg M, Sörensson P, Christersson C, Nielsen NE, et al. High incidence of infective endocarditis in adults with congenital ventricular septal defect. *Heart;* 2016. pii: heartjnl-2015-309133
13. Lee M, Chang S, Choi S, et al. Clinical features of right-sided infective endocarditis occurring in non-drug users. *J Korean Med Sci.* 2014;29:776-81.
14. Birkenkamp K, Jin J, Shivashankar R, Jouni H, Baddour L, Blauwet L. Ventricular septal defect and bivalvular endocarditis. *Avicenna J Med.* 2015;5:21-3.
15. Wang T, Oh T, Voss J, et al. Characteristics and outcomes for right heart endocarditis: Six-year cohort study. *Heart Lung Circ.* 2014;23:625-7.
16. A. Marc Gillinov, Ramon Diaz, Eugene H. Blackstone, Gosta B. Pettersson, MD, Joseph F. Sabik, Bruce W. Lytle, Delos M. Cosgrove III, Double Valve Endocarditis *Ann Thorac Surg.* 2001;71:1874 –9.
17. Pais JP, Sousa M, Mota R, Cambão AR, Nascimento A. Right-Side Endocarditis: A Typical Presentation in an Atypical Patient. *Cureus.* 2021;13:e18897.
18. Iversen K, Ihlemann N, Gill SU, Madsen T, Elming H, Jensen KT, Bruun NE, Høfsten DE, Fursted K, Christensen JJ, Schultz M, Klein CF, Fosbøll EL, Rosenvinge F, Schønheyder HC, Køber L, Torp-Pedersen C, Helweg-Larsen J, Tønder N, Moser C, Bundgaard H. Partial oral versus intravenous antibiotic treatment of endocarditis. *N Engl J Med.* 2019;5:415-424.
19. Randi E. Durden, Joseph W. Turek, Benjamin E. Reinking, Manish Bansal.

- Acquired ventricular septal defect due to infective endocarditis. *Pediatr Card.* 2018; 11:100-2.
20. Di Filippo S, Semiond B, Celard M, Sassolas F, Vandenesch F, Ninet J, Etienne J, Bozio A. Characteristics of infectious endocarditis in ventricular septal defects in children and adults. *Arch Mal Coeur V.* 2004;5: 507-14.

© 2022 Njie et al.; This is an Open Access article distributed under the terms of the Creative Commons Attribution License (<http://creativecommons.org/licenses/by/4.0>), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.