Cardiovascular and Metabolic Science www.ancam.org.mx

Clinical Case

Vol. 32 No. 2
April-June 2021

doi: 10.35366/99748

Isolated congenital left ventricular diverticulum in adult: an unusual finding in a cocaine-associated myocardial infarction

Divertículo ventricular congénito izquierdo aislado en adulto: un hallazgo inusual en infarto de miocardio asociado a cocaína

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Keywords:
Left ventricular diverticulum, isolated ventricular diverticulum, myocardial infarction, adult, coronary computed tomography angiography, cocaine use.

Palabras clave:
Divertículo ventricular izquierdo, divertículo ventricular aislado, infarto al miocardio, adulto, angiografía coronaria por tomografía computarizada, consumo de cocaína.

ABSTRACT

Congenital left ventricular diverticulum (CLVD) consists of an out-pouching of endocardium, myocardium, and pericardium, which often presents as a projection from the ventricular free wall, with a narrow neck connecting the cavity to the ventricle proper. Although it is often associated with other cardiac and extracardiac congenital anomalies, it may also present alone, as an incidental finding in adult patients. Due to its low overall prevalence and variability in presentation, a standardized treatment has yet to be delineated. We present the case of an adult patient with cocaine-associated myocardial infarction (MI), in which a septal CLVD was revealed by coronary computed tomography (CT) angiography. Other cardiac anomalies were ruled out and, the patient responded well to medical treatment after cardiac catheterization; thus, medical follow-up was preferred and did not surgery.

RESUMEN

El divertículo ventricular congénito izquierdo (DVCI) consiste en una bolsa de salida de endocardio, miocardio y pericardio, que a menudo se presenta como una proyección de la pared libre del ventrículo, con un cuello estrecho que conecta la cavidad con el ventrículo propiamente dicho. Aunque a menudo se asocia con otras anomalías congénitas cardiacas y extracardiacas, también puede presentarse solo, como un hallazgo incidental en pacientes adultos. Debido a su baja prevalencia general y variabilidad en la presentación, aún no se ha definido un tratamiento estandarizado. Presentamos el caso de un paciente adulto con infarto de miocardio (IM) asociado a cocaína, en el que se evidenció un DVCI septal mediante angio-TC coronaria. Se descartaron otras anomalías cardiacas y el paciente respondió bien al tratamiento médico tras el cateterismo cardiaco; por lo que se prefirió el seguimiento médico y no la cirugía.

INTRODUCTION

Congenital left ventricular diverticulum (CLVD) consists of an out-pouching of endocardium, myocardium, and pericardium, which often presents as a projection from the ventricular free wall, with a narrow neck connecting the cavity to the ventricle proper.[1] Although it is often associated with other cardiac and extracardiac congenital anomalies, it may also present alone, as an incidental finding in adult patients.[2] Due to its composition of normal cardiac tissue, as compared to a congenital left ventricular aneurysm (CLVA), the diverticulum contracts synchronously with the ventricle's remainder. Thromboembolic events, ventricular arrhythmias, heart failure, and free wall rupture have been documented to occur. Due to its low overall prevalence between 0.02% and 0.76%, and variability in presentation, a standardized treatment has yet to be delineated.[3] We present the case of an adult patient with cocaine-associated myocardial infarction (MI), in which a septal CLVD was revealed by coronary computed tomography (CT) angiography.

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How to cite: Pedraza-Jiménez R, Alanís-Naranjo JM, Morelos-Guzmán M. Isolated congenital left ventricular diverticulum in adult: an unusual finding in a cocaine-associated myocardial infarction. Cardiovasc Metab Sci. 2021; 32 (2): 105-109. https://dx.doi.org/10.35366/99748

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Received: 23/11/2020
Accepted: 03/02/2021

CASE PRESENTATION

A 42-year-old male with a history of cocaine use presented to the hospital with progressive retrosternal chest pain, diaphoresis, and nausea. He associated symptoms after «snorting» approximately 3 grams of cocaine over the last two days.

On examination, his heart rate was 115 bpm. There was no respiratory distress with normal heart and respiratory sounds. The initial 12-lead electrocardiogram (ECG) showed an inversion of T wave in leads V2-V5 with an elevation of ST-segment of 2 mm in leads V2-V4 *(Figure 1)*. Blood tests were notable for elevated troponin of 16,000 ng/L. A transthoracic echocardiogram revealed akinesia in the apical segments, a left ventricular ejection fraction of 35%, and a pericardial effusion of 3 mm.

A coronary CT angiography with dipyridamole stress test produced a calcium score of 0 Agatston Units; however, it revealed a 10 mm length non-calcified occlusive plaque at the proximal portion of the left anterior descending artery and pericardial effusion at right cavities *(Figure 2)*.

Also, CT revealed a left ventricular out-pouching of 1.8 × 2 cm (longitudinal and transverse diameter, respectively). A neck diameter of 6 mm, arising from the septal wall, adjacent to the occlusive plaque with contraction relative to the left chamber *(Figure 3)*.

Based on these findings, he was diagnosed as having a cocaine-associated MI and CLVD. The patient underwent cardiac catheterization, with the left anterior descending artery's immediate blood flow after balloon dilation *(Figure 4)*. Medical treatment controlled his symptoms, and he was discharged symptom-free. He continued follow-up at cardiology and cardiothoracic surgery units.

At the one and two-year follow-up, the patient continued asymptomatic. Follow-up echocardiograms showed a left ventricular ejection fraction of 65% with no pericardial effusion and CT with no changes in the size of the CLVD.

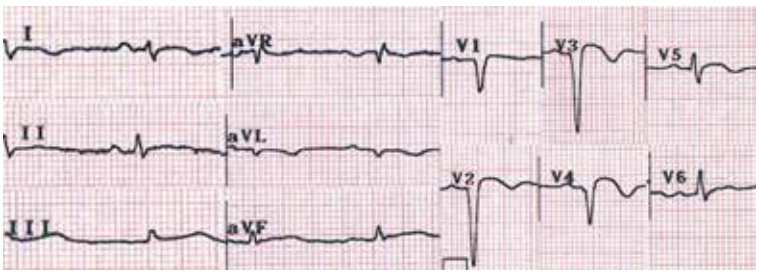

Figure 1: Initial 12-lead electrocardiogram: inversion of T wave in leads V2-V5 with an elevation of ST-segment of 2 mm in leads V2-V4 (anteroseptal necrosis).

DISCUSSION

CLVD is a rare and usually asymptomatic cardiac malformation. It frequently accompanies other cardiac abnormalities, and it is most commonly diagnosed during early childhood. About 30% of all cases are not associated with a congenital malformation, and they are defined as isolated CLVD. It is frequently challenging to diagnose CLVD because of its asymptomaticity.[4]

The differential diagnosis of left ventricular out-pouchings includes aneurysm, pseudoaneurysm, and diverticulum. According to Marijon et al., CLVD and aneurysm represent two distinct entities with different histological and morphologic characteristics and outcomes. The diverticulum is characterized by synchronal contractility and three myocardial layers on histological examination. There are two subtypes of CLVD: apical type cases are always associated with other cardiac anomalies or midline thoracoabdominal defects, whereas non apical type cases occur in isolation.[5]

According to structural characteristics of the wall, CLVD can be classified as muscular or fibrous. Fibrous CLVD consists mainly of fibrous tissue with few or no muscle fibers. The muscular type of CLVD must be distinguished from LV noncompaction, characterized by a prominent trabecular meshwork with a distinctly spongy appearance and deep intertrabecular recesses believed to be caused by an arrest in normal embryogenesis. When the CLVD is associated with congenital anomalies of the thoracic and abdominal midline, diaphragmatic and sternal defects, and partial absence of the inferoapical pericardium, this scenario is called syndrome or pentalogy of Cantrell.[4] CLVA represents the other end of the spectrum, whereby the out-pouching is dyskinetic or altogether akinetic

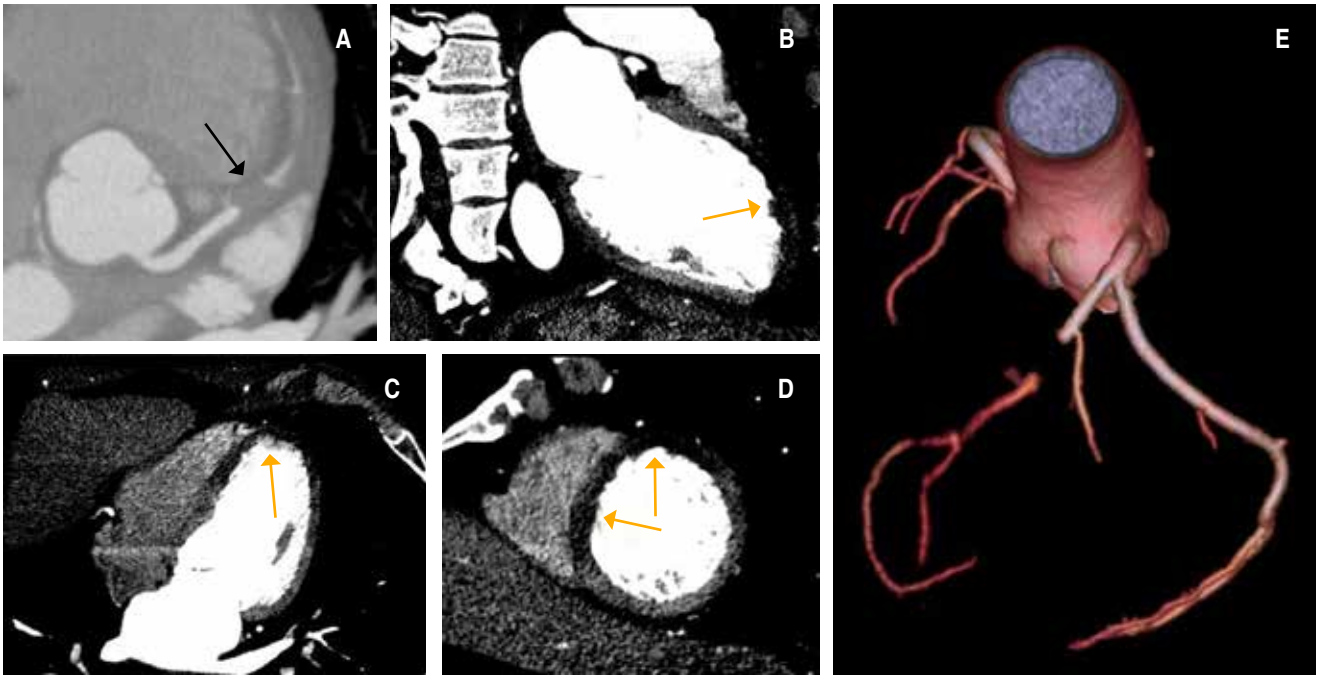

Figure 2: Coronary computed tomography angiography. **A)** Non-calcified occlusive plaque in the proximal third of the left anterior descending artery (arrow). **B)** Two-chamber view: perfusion defect in the anterior wall during the stress test (arrow). **C)** Four-chamber view: perfusion defect in the septal and lateral wall at middle and apical segments (arrow). **D)** Two-chamber view: perfusion defect in the anterior, anteroseptal and inferoseptal surfaces at the mid-segment (arrow). **E)** 3D reconstruction of coronary arteries: Total occlusion of the left anterior descending artery.

and histologically composed of fibrous tissue, dissimilar to that of the ventricular wall.[6]

CLVD is often clinically silent but may be associated with systemic embolism, arrhythmias, heart failure, myocardial ischemia, and cardiac rupture.[7] There are few reports in the literature of an isolated CLVD presenting as myocardial ischemia. However, this patient was asymptomatic for many years and presented to the hospital with chest pain due to cocaine-associated MI.

Mechanisms of acute MI resulting from cocaine use are multifactorial. At low doses, cocaine-induced sympathetic effects increase heart rate, blood pressure, and myocardial contractibility, leading to increased myocardial oxygen demand. Cocaine also enhances coronary spasm/vasoconstriction and platelet adherence/thrombosis, leading to the reduced myocardial oxygen supply. Thus, an imbalance between oxygen supply and demand results in MI. At high doses, cocaine-induced local anesthesia results in decreased left ventricular contractibility and prolonged QRS and QT intervals in electrocardiograms by blocking sodium transport and norepinephrine uptake in the myocardium. In vessels, cocaine contributes to MI by increasing endothelin-1 and reducing nitric oxide production in endothelial cells. When vessels are stressed, acute damages/ruptures can occur, promoting thrombosis by increasing platelet activity/aggregation and elevating fibrinogen levels and plasminogen activator inhibitor activity. These cellular and molecular cascades result in reduced cardiac blood flow, leading to acute MI and possibly atherosclerosis and coronary thrombosis in the long term. As such, cocaine induces acute MI by directly affecting myocardial tissues in the heart and indirectly enhancing thrombosis in vessels.[8] In this patient, the abnormal distribution of vessels due to septal CLVD and the effects of cocaine use could be the triggers of the myocardial infarction.

Diagnosis of CLVD can be made by echocardiography, computed tomography

angiography, magnetic resonance imaging, and cineangiography.[7] Ventriculography (95.5%), CT (88.9%), CMR (84.2%), and echocardiography (78.2%) are all sensitive tools for diagnosing CLVD.[9] Because of the patient´s clinical features, ECG, and enzyme changes, it was felt at first that this represented a cocaine-associated MI. For further evaluation, transthoracic echocardiography, coronary CT angiography, and cardiac catheterization were performed. Transthoracic echocardiography showed no ventricular pouch. Coronary CT angiography revealed abnormal coronary arteries with a left ventricular out-pouching arising from the septal wall. The pouch showed contraction relative to the main chamber, as the findings reported in ventricular diverticulum cases.

The incidence of adverse events in symptomatic patients with CLVD is increased during long-term follow-up.[7] In a review of 809 patients published since 1816 with either CLVD or CLVA, 4.5% of patients with CLVD were found to have an episode of rupture, and a 5% reported cardiac death rate. Other significant complications included ventricular tachycardia/fibrillation in 13.1%, embolic events 3.6%, and syncope 5.1%.[10]

Because of the inadequate data for universal guidance, management of CLVD remains unclear, and treatment options include close observation and surgery. Surgical removal should be considered based on the localization of the lesion and associated symptoms. Unfavorable anatomy of the diverticulum and patient's reluctance to undergo surgery adjudicated in favor of conservative management.[11]

Despite a concise review of the cases reported to date, there is little in the way of a detailed surgical approach. Some authors reported closure of the diverticulum neck with a patch, surgical glue closure, and plication with aneurysmorrhaphy. Other authors have described their technique as suture reapproximation in a double-layered fashion with the use of felt. In contrast, some describe the removal of the out-pouching cavity and closure with an *in-situ* patch. In 2020, Mejia J et al. reported a successful repair of a CLVD in an adult using a two-patch technique.[3]

Poor prognoses, including high mortality and morbidity, have been reported in infants and children with comorbid defects, and serious complications associated with CLVD include arrhythmias, embolism, cardiac failure, and rupture. In contrast, more favorable clinical outcomes have been demonstrated, most notably in asymptomatic adult patients, which may depend on no significant comorbidity.[12] In this case, other cardiac anomalies were ruled out, and the patient responded well to medical treatment after cardiac catheterization; thus, medical follow-up was preferred and did not surgery.

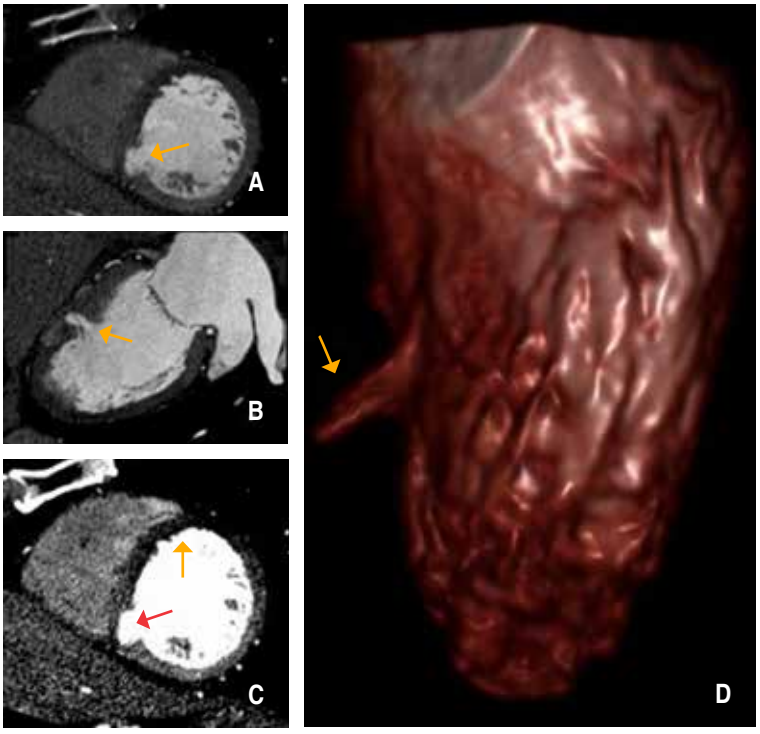

Figure 3: Coronary computed tomography angiography revealing a left ventricular diverticulum. **A & B)** Two-chamber view (arrow). **C)** Two-chamber view: Perfusion defect in the anterior and anteroseptal wall at the mid-segment (arrow). Ventricular diverticulum adjacent to ischemic myocardial tissue (red arrow). **D)** 3D reconstruction of left ventricular volume (arrow).

CONCLUSIONS

We present a patient with a rare cardiac malformation, with few cases documented in the literature. CLVD can simulate various heart diseases, and early diagnosis would be difficult due to a nonspecific clinical presentation.

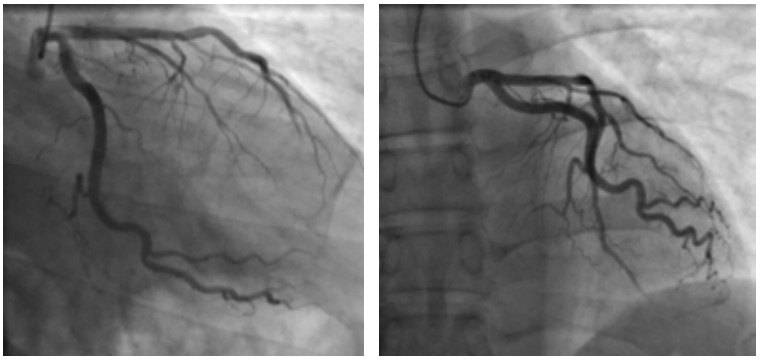

Figure 4: Cardiac catheterization showing blood flow of the left anterior descending artery after balloon dilation.

There are few reports in the literature of an isolated CLVD presenting as myocardial ischemia. The abnormal distribution of the coronary vessels due to septal CLVD and the effects of cocaine use could be the triggers of myocardial infarction in this patient. This case showed the importance of the cardiovascular imaging approach for the diagnosis of CLVD. Management remains unclear, and treatment options include close observation and surgery. Surgery in adults with isolated CLVD could be considered in symptomatic patients. The risks and prognosis of this malformation are challenging to assess due to its extremely low incidence. More studies are needed to determine the appropriate treatment.

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Funding/support: This study did not benefit from any funding organization in the public or commercial.

Conflict of interests: The authors declare no conflict of interest.

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