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Percutaneous transcatheter device closure of a congenital left ventricular diverticulum in a 5-year-old child

Fechamento transcateter de divertículo ventricular esquerdo congênito com dispositivo percutâneo em criança de 5 anos

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ABSTRACT – Ventricular diverticulum is a rare congenital malformation. Although most patients are asymptomatic, it can present with rupture and sudden death, for which surgical repair is indicated. The authors report the case of a 5-year-old boy with a prenatal diagnosis of an isolated left ventricular diverticulum. It was decided for surgical closure; however, a persisting leakage at the patch repair site was observed, for which a transcatheter percutaneous closure approach was used, achieving complete occlusion of the defect. Transcatheter closure of suitable ventricular diverticula is a safe and effective option.

Keywords: Heart defects, congenital; Heart ventricles/abnormalities; Diverticulum/congenital; Cardiac catheterization/adverse effects; Septal occluder device

RESUMO – O divertículo ventricular é uma malformação congênita rara. Embora a maioria dos pacientes seja assintomática, podem ocorrer ruptura e morte súbita, sendo por isso indicada a correção cirúrgica. Os autores relatam o caso de um menino de 5 anos com diagnóstico pré-natal de divertículo ventricular esquerdo isolado. Foi decidido realizar um fechamento cirúrgico. No entanto, observou-se fluxo persistente no local do retalho para correção, para o qual foi utilizada abordagem de fechamento percutâneo transcateter, obtendo-se a oclusão completa do defeito. O fechamento transcateter de divertículos ventriculares adequados é uma opção segura e efetiva.

Descritores: Cardiopatias congênitas; Ventrículos cardíacos/anormalidades; Divertículo/congênito; Cateterismo cardíaco/efeitos adversos; Dispositivo para oclusão septal

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INTRODUCTION

Ventricular diverticulum is a rare congenital cardiac malformation.[1] It can present as an isolated defect or as part of a pentalogy described by Cantrell et al.[2] The prevalence varies between 0.02% and 0.76%.[3,4] Most patients are asymptomatic, and diagnosis is usually made incidentally.[5] However others may present with chest pain, ventricular arrhythmias, systemic embolisms, rupture of the ventricular wall and sudden death.[6] There are no guidelines for the management of this entity. Surgical approach is usually performed.[7] Percutaneous management in children has not been widely described in the literature. Only one case report was found that performed closure of the defect with a device for closure of patent ductus arteriosus.[8]

The purpose of this case report was to present the percutaneous transcatheter device closure of a ventricular diverticulum in a five-year-old boy, who presented a residual leak after surgical repair of the defect.

CASE REPORT

A 5-year-old child with native anatomy of a left ventricular aneurysm/diverticulum diagnosed in the prenatal evaluation. At birth, he had spontaneous neonatal adap-

tation with subsequent respiratory distress, which required admission to the neonatal intensive care unit. He was transferred to the pediatric cardiology service at 19 days of age to determine the management. A transthoracic echocardiography was performed, which revealed aneurysm of the distal third of the left ventricle (LV) free wall and of the entire apex, measuring 1.5cm x 2.5cm in diameter, and severe hypokinesia of the aneurysmal area. The heart team decided to perform a diagnostic cardiac catheterization to evaluate the anatomy of the cardiac defect. At 6 months of age, he was taken to the Cath Lab. Left ventriculography showed a giant diverticulum, with trabeculae inside and associated hypokinesia in the apical segment of the LV. Since the patient remained asymptomatic, a conservative management was proposed, and he stayed in close follow-up with the heart team until 2 years of age. A cardiac magnetic resonance imaging was performed and showed persistence of the defect; hence, surgical approach was decided. At 4 years and 5 months of age he was taken to surgery, when the diverticulum was repaired with a heterologous pericardial patch. The procedure did not present any complications, he was taken to the intensive care unit for postoperative care, where he remained for 5 days and was subsequently discharged.

Twenty-seven days after the surgical procedure, a control transthoracic echocardiography was performed, revealing the presence of a residual pericardial bag at the apex of the ventricle, and two residual leaks with bidirectional shunting (Figure 1). The case was discussed with the heart team, and it was decided to perform endovascular closure of the defect. At 5 years and 2 months of age, he was taken to the Cath Lab. Left ventriculography was performed and the injections showed a giant diverticulum with a tunnel-like proximal neck in the apical segment of the LV (Figure 2). To clarify the anatomy, a 10mm x 20mm Tyshak balloon was advanced and inflated manually, observing that the neck of the diverticulum measured 4mm. It was decided to occlude the defect with a 10mm Amplatzer™ Vascular Plug II (AVP II) device. A 6F flexor introducer was passed to the LV, then the distal non-taper catheter was advanced into the diverticulum and a 0.035 Amplatz support guide was passed. A JR 6F guide catheter was passed over this, and a 10mm AVP II device was advanced later, releasing the distal end and the central drum in the diverticulum cavity and in the distal neck. Finally, the proximal disc was released. Control angiography showed the device in position with occlusion of the diverticulum (Figure 3). No complications were reported. A transthoracic echocardiography on the next day showed a residual pericardial pocket occluded by the device.

The patient was prescribed enoxaparin, aspirin and warfarin for 3 months, and was admitted for the anticoagulation program. He was discharged and remained asymptomatic at 2 weeks follow-up.

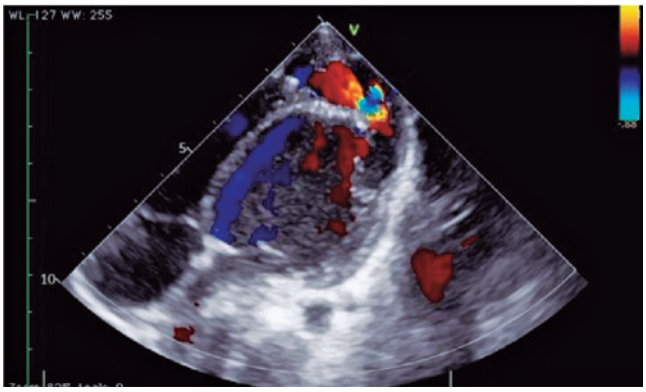

Figure 1. Transthoracic echocardiographic images showing the surgical patch and a leak at the end of the patch as demonstrated by color Doppler.

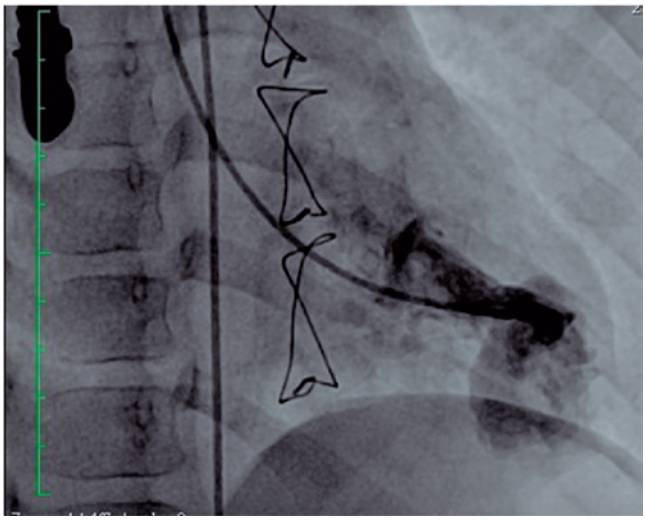

Figure 2. Lateral view of the angiogram showing the ventricular diverticulum with a narrow neck.

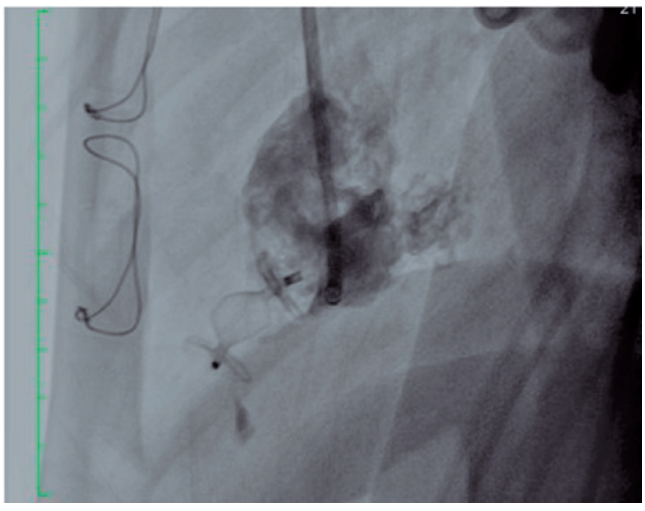

Figure 3. Control angiography images showing the device in position with occlusion of the diverticulum.

DISCUSSION

Ventricular diverticula are rare congenital cardiac malformations that consist of a protrusion of the free wall of the ventricle. They develop in the fourth embryological week, and it is believed that their formation can be explained by a partial arrest in the development of the embryonic ventricle at this stage.[4] It is difficult to differentiate between a congenital diverticulum and a congenital aneurysm. Diverticula are usually shaped like a finger or hook, they connect to the ventricular cavity by a narrow junction, and contract simultaneously with it; they can be associated with thoracoabdominal midline defects and other cardiac malformations, such as those described in the Pentalogy of Cantrell et al.[2] In contrast, the ventricular aneurysm is characterized as an akinetic or dyskinetic protrusion of the free wall of the ventricle, and communicates extensively with it.[1] Diverticula can generally be found in the LV; they can also be associated to the right atrium, right ventricle, and can be biventricular.[9]

The incidence is not very clear, since there are no precise data in the literature. A retrospective echocardiographic study described 16 cases of patients with diverticula or ventricular aneurysms in a population of 43 thousand adults, with a prevalence of 0.04%. Another study carried out on 12,924 autopsies of children found 750 cases of congenital cardiac malformations, and only three of them presented a diagnosis of ventricular diverticula, resulting in a prevalence of 0.02%.[4]

The clinical presentation varies. In the prenatal period it can generate pericardial effusion and arrhythmias,[10] in childhood most patients are asymptomatic, and diagnosis is made incidentally during other diagnostic procedures. An analysis of 809 cases of aneurysms and ventricular diverticula, carried out by Ohlow et al., showed that 2.9% of patients evaluated had a history of an embolic event, 4.2% presented with rupture of the ventricular wall, 19.7% presented syncope and heart rhythm disorders, chest pain was reported in 16.6% and signs of congestive heart failure in 6.8%.[7] In the case of our patient, postpartum evaluations revealed mild biventricular systolic dysfunction and mild ventricular extrasystole, with an arrhythmic load of 0.6%.

Transthoracic and transesophageal echocardiography continue to be important tools in evaluation of these malformations; however, they can be overlooked due to the difficulty in visualizing the ventricular apex with this technique. The gold standard continues to be invasive angiography, for it demonstrates the exact location of the defect, its communication with the ventricular chamber, its relation with the mitral valve apparatus and the coronary arteries, and its contractile nature. Cardiac tomography and cardiac magnetic resonance imaging can also be performed as in the case of our patient; however, this technique is not always available, and its relevance has not been defined in the literature yet.[4] Regarding the histological study, in the case of the diverticulum the three layers of the ventricular wall can be observed, including migration of the coronary artery and preservation of myocardial architecture.[11]

There are no specific guidelines for the management of ventricular diverticula. The study carried out by Ohlow et al. found that in 45.3% of cases surgery was performed to repair the diverticulum, and simultaneously to correct other associated cardiac malformations.[7] The conservative strategy with close follow-up in cases of an isolated diverticulum, with no other associated malformations, can also be performed. Nonetheless, the diverticulum has the potential to cause embolisms, arrhythmias, heart failure and rupture; all these justify the recommendation of surgical correction after making diagnosis.[12]

The field of percutaneous correction for congenital heart disease has recently experienced great development. To our knowledge, there is only one case report of a transcatheter closure of a ventricular diverticulum. This was carried out by Jain et al., in a 12-year-old patient, who had an isolated diverticulum in the LV. She was taken to the Cath Lab and LV angiography was performed and determined the size of the diverticulum was 20mm x 12mm with a long neck 6mm in diameter. Percutaneous transcatheter closure was performed with a patent ductus arteriosus occlusion device. Subsequent angiogram showed minimal residual flow through the device, and echocardiography the following day found the device in proper position. The patient was discharged and prescribed aspirin and remained asymptomatic after 24 months of follow-up. In this report, the authors emphasized that many diverticula may not have a short neck within their structure; hence, not all of them are susceptible to intervention through cardiac catheterization.[8]

With this report, it can be recognized that isolated ventricular diverticula are susceptible to percutaneous correction as an alternative to surgery, with lower morbidity and mortality.

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None.

CONFLICTS OF INTEREST

The authors declare there are no conflicts of interest.

CONTRIBUTION OF AUTHORS

Conception and design of the study: ML; data collection: ML; data interpretation: RLV, LHD and RC; text writing: RLV, LHD and RC; approval of the final version to be published: RLV.

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