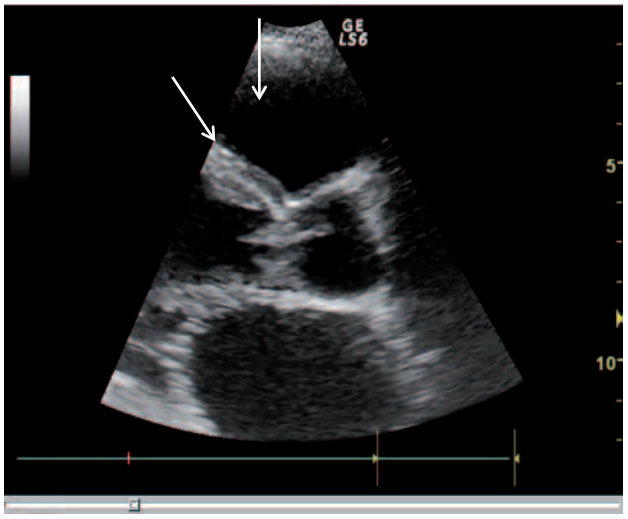

Fig. 2. Transthoracic echocardiography image, zoom of left parasternal axis. The arrows show vegetation in the aortic and mitral valves.

Conflicts of interest

None declared.

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

Soledad Orazi, Hugo Alberto Gay, Emiliano Torres, Daniel Esteban Calfunao, Claudia Patricia López, María Rosa Núñez

Department of Cardiology, Department of Infectology, Department of Bacteriology, and Department of Epidemiology. Hospital Provincial de Neuquén "E. Castro Rendón"
Buenos Aires 445, Neuquén Capital, Pcia. de Neuquén, Argentina
e-mail: cardiologiahpn@gmail.com

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Congenital Fistula Between the Internal Mammary Artery and the Pulmonary Trunk

Congenital internal mammary artery to pulmonary artery fistula is a rare condition, and its diagnosis is exceptional in pediatric patients. (1) The first case of mammary artery to pulmonary artery fistula was published in 1947, (2) and very few cases have been described since. This rare condition can be either congenital (it occurs in 1 out of 50,000 patients with congenital heart disease) or acquired (usually secondary to coronary artery bypass surgery, traumas, inflammation, or neoplasia), (3) and its diagnosis is exceptional in patients with no evidence of disease or triggering factors. (4)

Congenital forms are associated to pulmonary atresia or tetralogy of Fallot among other heart diseases, and to pulmonary sequestration and arteriovenous malformations. The embryonic origin of these connections is not well known, although it is believed that in these cases, systemic-pulmonary fistulas are formed when the main pulmonary arterial system does not develop continuity with the embryonic lung and cannot form a normal pulmonary arterial tree; (4) this is supported by the common embryonic origin of the chest wall and the pulmonary tree. Several authors have suggested that congenital fistulas arise because pulmonary capillary vessels and the aorta, which connect systemic and pulmonary circulation in the fetus, fail to regress. (5)

In general, patients with congenital fistulas but no other associated anomalies are asymptomatic. It is diagnosed in a study to detect heart murmur, although its clinical presentation depends partly on the functional repercussion of the fistula, which will be proportional to the size of the implicated vessels, and where it is located in relation to the heart or drainage site.

With time, fistulas may cause vessel dilation and symptoms such as congestive heart failure, bacterial endocarditis and/or rupture. Treating this condition is controversial and the options are an expectant attitude, percutaneous closure, or surgery. (2)

We describe the case of a 10-year old boy with no relevant medical history, who was referred for heart murmur evaluation. Physical examination showed a continuous heart murmur at the left superior sternal border, with no signs of heart failure. The electrocardiogram showed no repolarization changes or other anomalies. Doppler-echocardiography detected diastolic flow suggestive of fistula at the pulmonary trunk, without coronary artery dilation or anomalies or evidence of the origin of such drainage. No other structural abnormalities or change in the size of heart chambers were targeted. Although the initial suspicion was of coronary artery fistula, the evaluation was completed with a contrast computed tomographic angiography, which revealed an anomalous vessel with a tortuous path at the level of the internal

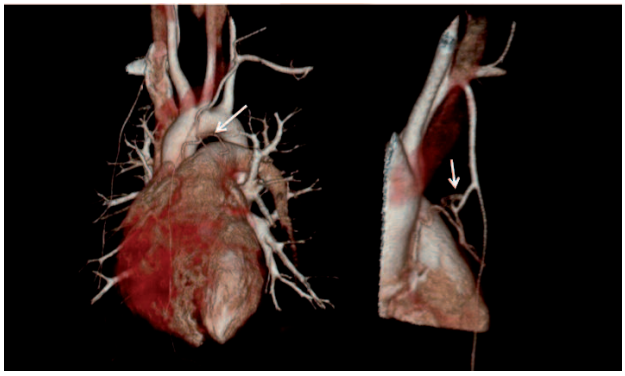

Fig. 1

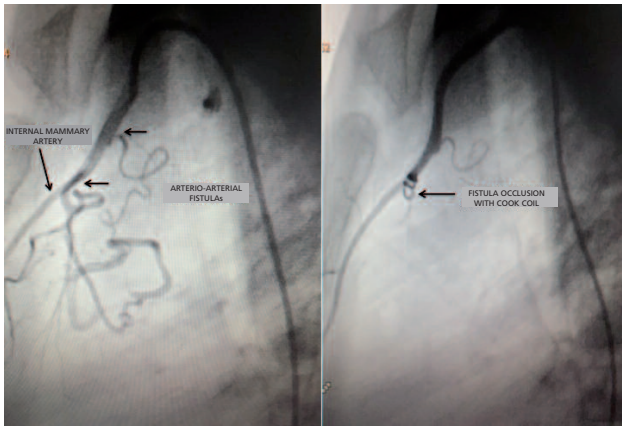

Fig. 2

mammary artery draining in the main pulmonary artery (Figure 1). Further angiographic evaluation was decided, which confirmed the fistula and another one of smaller caliber in the immediately upper portion of the internal mammary artery. Despite the patient was asymptomatic, given that the fistula size was significant and could cause complications in the future, uneventful embolization of the larger vessel was successfully performed with a Cook coil (Figure 2). The patient has remained asymptomatic and without further complications.

Although coronary artery fistulas are quite common anomalies, they are difficult to differentiate from those originated at another level, as is the case of this arterio-arterial fistula with greater potential risk for long-term complications. Therefore, when in doubt, diagnosis should be completed with other imaging techniques or with angiography, which is useful to determine the size, location, and shunt hemodynamics.

When the patient is asymptomatic and presents no other disorders, treating this condition is controversial because its progression is unknown due to the limited caseload. Some authors choose the expectant attitude approach, although in most cases surgery is the option due to its positive outcome and low morbidity and mortality. (2) Percutaneous closure of these defects is relatively recent, the case described by Fernández et al. in 2004 using a mixed technique with an Amplatzer Duct Occluder (ADO) associated to coils, being the first one described in the literature. (2) Since then, other authors have performed percutaneous closure with stents or fluid embolization with N-butyl cyanoacrylate with positive outcomes in adult patients.

Although congenital isolated arterio-arterial mammary artery-to-pulmonary artery fistula is a rare condition, percutaneous closure can be useful in pediatric patients as demonstrated in our case.

Conflicts of interest

None declared.

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Ariadna Ayerza Casas[1], Daniel Palanca Arias[1], Marta López Ramón[1], Elena G. Corella Aznar[1], Cristina Gutierrez Alonso[2], Lorenzo Jiménez Montañés[1]

[1] Pediatric Cardiology Division. Department of Pediatrics.
[2] Pediatric Radiology Division. Department of Radiology.
Hospital Universitario Miguel Servet, Zaragoza. Spain.
email: aayerzac@hotmail.com

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