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Case Report

Surgical Repair Following Aorto-to-Right Atrial Fistula Transcatheter Closure: A Case Report

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Abstract

Aorto-to-Right Atrial fistula (ARAF) is a frequent complication of ruptured Aortic Sinus of Valsalva aneurysm, traditionally requiring surgical treatment. Endovascular closure is a management option nowadays available with good clinical outcomes. In this report, we present the singular case of a recurrent ARAF, sixteen years after transcatheter closure. At the admission at our department, the Patient presented with symptoms and signs of acute heart failure. Elective surgical repair was the treatment of choice.

Introduction

Sinus of Valsalva aneurysm is a rare cardiac anomaly, frequently affecting the right coronary sinus (70-90%) [1]. Ruptured sinus of Valsalva aneurysm typically leads to the formation of a fistula with the right atrium or ventricle, resulting in a severe left-to-right shunt. Surgical repair should be promptly considered since the right ventricular overload would progressively deteriorate the heart function. However, in the last decades, transcatheter closure emerged as an attractive alternative method for repair [2]. At present, whether to perform the former or the latter approach remains a challenging question, being hints to its solution provided by the statistics of case reports and series. Here we describe the surgical repair of a recurrent Aorto-to-Right atrium fistula (ARAF), sixteen years after the endovascular management, aiming at enriching scientific literature on this topic.

Case

A 55-year-old woman, with a recent history of dyspnea at rest, presented for evaluation at our institution. Past medical history included: permanent atrial fibrillation, hypothyroidism, thalassemia trait, and a percutaneous catheter closure of an ARAF, secondary to a ruptured Non-Coronary (NC) Sinus of Valsalva aneurysm. It was performed sixteen years earlier at another institution. On that occasion, the first attempt to place an Amplatzer duct occluder 8/6 mm, was unsuccessful. Because of a premature screwing, the device embolized in a peripheral branch of the pulmonary artery, and unsuccessful attempts were made to retrieve it. An Amplatzer duct occluder 10/8 mm was finally located, without residual left-to-right shunt [3]. On admission to our department, her functional capacity was New York Heart Association (NYHA) class IV [4]. During the previous months, she reported the rapid onset of worsening shortness of breath, impaired exercise tolerance, and 10kg of weight gain. Physical examination showed continuous grade 4/6 murmur well heard along with all cardiac auscultations sites, pulmonary crepitation, associated with peripheral pitting edema, and elevated jugular venous pulse. Chest X-ray revealed bilateral basal pleural effusion. Transthoracic echocardiography showed the presence of left to right shunt associated with right atrium enlargement and severe pulmonary hypertension, without structural defect of the aortic valve and alteration of the left ventricular function. These data were confirmed at the cardiac catheterization study. A hole of communication between the cranial portion of the aneurysmatic NC sinus of Valsalva and the right atrium, near the Amplatzer device, was documented at the ascending aortography. Baseline Pulmonary Artery Pressure (PAP) was 103/68 mmHg (mean 58 mmHg), halved after O2 administration (mean 36 mmHg). The calculated pulmonary to systemic flow ratio (Qp:Qs) was 2:1. No abnormalities of the coronary arteries were reported. The Patient

underwent an elective open surgical repair of the ARAF via a median sternotomy. Cardiopulmonary bypass strategy included: bicaval and ascending aorta cannulation, left ventricular venting, antegrade and retrograde Buckberg cardioplegia. After the right atriotomy, specific attention was paid to remove the Amplatzer device as a single piece. Bovine pericardium was used to patch the NC sinus and the right atrial wall with a continuous suture. We performed a partial aortotomy to control the correct closure of the fistula, not reporting any residual leak. The aortic wall was then sutured, and a further dose of antegrade cardioplegia was administered to confirm the correct repair of ARAF from the right atrial view. CPB and aortic cross-clamp times were 153 and 105 minutes, respectively. The postoperative period was uneventful. Pre-discharge transthoracic echocardiogram confirmed the absence of Aorto-Atrial shunt, with moderate pulmonary hypertension (PAPs 50-55 mmHg) (Figures 1 and 2).

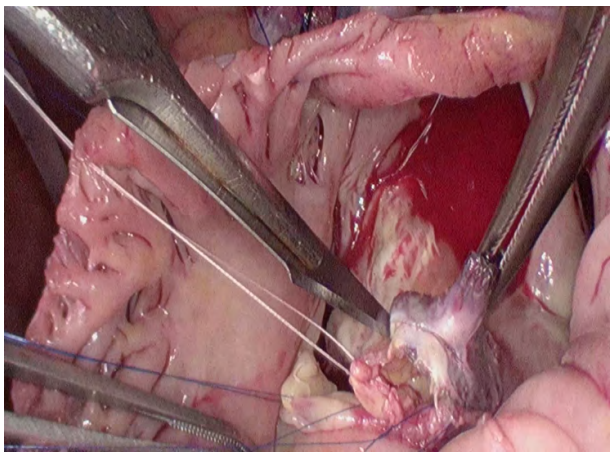

Figure 1: Operative view looking down into the right atrium. The Amplatzer occluder was removed as a single piece.

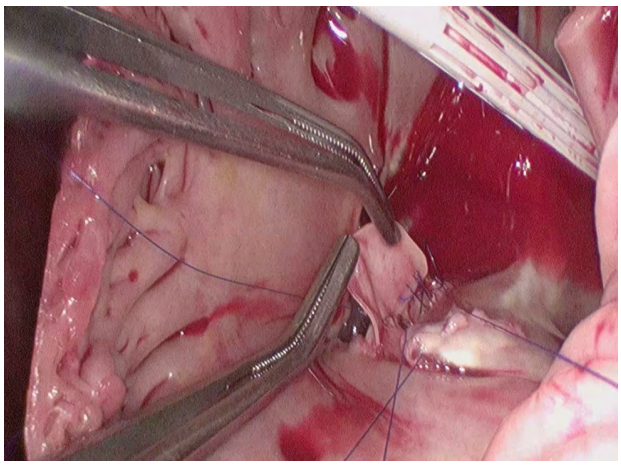

Figure 2: Operative view from the right atrium showing the ARAF repair using a bovine pericardium patch.

Discussion

Aorto-Atrial fistula is an uncommon but potentially life-threatening condition. It may be congenital or secondary to a wide range of diseases, including infective endocarditis and aortic dissections, or iatrogenic in nature [5]. When congenital, fistulas have been associated with genetic and connective tissue disorders and hypertension. There are a few cases of NC sinus aneurysms described in the scientific literature. Presenting the septal wall of the right atrium, its rupture in this chamber may occur with ARAF formation. Due to the low incidence of ARAF and the absence of clinical trials, therapeutic strategies rely on expert opinions and data available from case reports and case series on this topic. Surgical repair is traditionally recognized as the standard approach. Since the primary surgical closure was associated with a higher rate of recurrence [6], patch repair has rapidly become the preferred surgical strategy with optimal early and long-term follow-up outcomes. Akazie et al reported a case series of 34 patients who underwent surgical repair for congenital ARAF, with a survival rate of 90±7% and freedom from reoperation of 83±9% at 10 years [7]. Re-interventions were mainly correlated with primary rather than patch closure and the late development of aortic insufficiency. In the last decades, multiple endovascular procedures emerged as an attractive alternative to surgical repair. Given the absence of a specific device, Amplatzer occluders [8], balloon closures, and coil embolization were adapted for this purpose [9]. A careful assessment of the aortic valve leaflet is mandatory before releasing the device. Moreover, possible device embolization into the pulmonary or general circulation should be also taken into account, given the impact on early morbidity and long-term prognosis of these Patients. A recent systematic review reported good results about early mortality after transcatheter closure, with the lack of data about long-term morbidity and mortality [9]. In this paper, we present the case of a recurrent AAF, sixteen years after the endovascular closure. Since the Patient did not follow regular controls, the time of recurrence was not clear. The underlying right-sided volume overload led to an increase in pulmonary pressure, worsened by the early peripheral embolization of the Amplatzer device. The interventional cardiologist deemed this patient no longer a candidate for percutaneous treatment and a successful surgical repair with a bovine patch was performed, with rapid improvement of pulmonary hemodynamics. According to our experience and the natural evolution of the disease, we believe that surgical closure with a pericardial patch is the safest and most effective strategy of repair, with a better long-term impact on hemodynamics and recurrence. Moreover, this case report highlights the key role played by regular clinical and instrumental follow-up, in the early detection of recurrence and pulmonary hypertension which negatively impacts the prognosis of these patients.

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