Surgical repair after ineffective device closure of an inferior sinus venosus defect
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INTRODUCTION

Catheter-based device closure has long become the first-line treatment modality for closure of oval fossa atrial septal defects (ASDs) [1, 2]. For intra-atrial communications with more complex morphologic features, however, surgical correction is usually necessary to achieve an optimal long-term result [3, 4]. We present a case of incomplete percutaneous closure of an inferior sinus venosus ASD that led to obstruction of the inferior caval vein and required mitral valve repair.

CASE PRESENTATION

A 21-year-old woman presented with a 4-year history of decreasing exercise capacity. At the age of 17 years, she had undergone percutaneous transfemoral closure of an ASD with an Amplatzer Septal Occluder® (32 mm) system in an external adult cardiology department. Her medical history also included hereditary factor V Leiden and intolerance for acetylsalicylic acid. At the time of admission, she had significant dyspnea and cyanosis during mild exercise, and experienced repeated episodes of chest pain and limb paresthesia. Electrocardiogram showed sinus rhythm (53 bpm) and incomplete right bundle branch block. Lung function test results were appropriate, maximum oxygen uptake was reduced (VO2max: 17–20 ml/kgBW) and peripheral oxygen saturation was 90–95% at rest and dropped to 70% during exercise. Pulmonary embolism was ruled out by computed tomography, and there was no evidence of pulmonary hypertension at rest or during exercise. In the echocardiogram, both right ventricular and left ventricular function was normal. The 32 mm Amplatzer device nearly filled the right atrium, restricting both the inflow of the inferior caval vein (IVC) and diastolic flow across the tricuspid valve. There was also residual left-to-right shunting at the caudal aspect of the device (38%). Upon right- and left-heart catheterization, the device was shown to be placed asymmetrically on the atrial roof with the lower edge completely sitting on the anterior orifice of the IVC, producing a pressure gradient of 5 mmHg (Fig. 1). The defect appeared to lack a rim towards the IVC orifice. In the left atrium, the device seemed to be in contact with the mitral valve but did not compromise its function.

Owing to the clinical symptoms, it was decided to explant the device surgically. After median sternotomy, total cardiopulmonary bypass was established and cardiopulmonary arrest was initiated. The right atrium was opened, and the device was found to be attached to the mitral valve tissue at the posterior wall of the left atrium that was closed by direct suture. The aortic cross-clamp was removed. After the termination of extracorporeal circulation, transesophageal echocardiography showed significant mitral valve regurgitation. ECC was re-established, the heart was arrested again and the left atrium was opened. Careful inspection of the mitral valve showed that the device had been attached to the mitral valve tissue at the posterior commissure, and that part of the posterior leaflet in segment P3 was required. This case emphasizes the importance of appropriate patient selection for interventional ASD closure and demonstrates that patients with less-than-optimal results should be referred for surgical correction early, before the device produces secondary damage to atrial structures and atrioventricular valves.

Abstract

A young woman presented with severe heart failure symptoms 4 years after percutaneous device closure of an atrial septal defect (ASD). There was residual left-to-right shunting, and the device was obstructing the inferior caval vein and tricuspid valve flow. Intraoperatively, the ASD was shown to be an inferior sinus venosus defect, and reconstruction of the mitral valve and the posterior wall of the left atrium was required. This case emphasizes the importance of appropriate patient selection for interventional ASD closure and demonstrates that patients with less-than-optimal results should be referred for surgical correction early, before the device produces secondary damage to atrial structures and atrioventricular valves.

Keywords: Atrial septal defect · Sinus venosus · Device
had been damaged during device explantation, which appeared to be competent by saline testing during the primary inspection. The commissure was reconstructed with several pericardium-enforced mattress sutures. The mitral valve ring at the P3 segment was plicated as in a unilateral Whooler plasty, and the posterior commissure was partially closed with a mattress suture, making sure that anterior mitral leaflet mobility was not significantly impaired. Now, mitral valve function was adequate and the operation was completed as usual. The postoperative course was uneventful. Transthoracic echocardiography at follow-up displayed an intact inter-atrial septum and normal atrioventricular valve function, and the clinical symptoms have completely disappeared.

**COMMENT**

Intra-atrial communications located within the oval fossa (patent foramen ovale and secundum ASD) are routinely closed using catheter-based devices, provided that sufficient tissue surrounds the defect circumferentially. Therefore, ostium primum ASDs (ASD I, incomplete atrioventricular canal) and superior as well as inferior sinus venosus defects need to be corrected surgically, although some interventional cardiologists still propagate device closure of complex ASDs [5]. In the case presented here, an inferior sinus venosus defect lacking a rim towards the IVC orifice was attempted to be closed with an oversized Amplatzer device, resulting in residual shunting at the caudal, rimless aspect, and in obstruction of IVC and tricuspid valve blood flow. Because 4 years had passed since the implantation of the device, it was firmly attached to the posterior left atrium wall as well as the posterior commissure of the mitral valve, so that surgical reconstruction of these structures was necessary. Surgery for failed device placement is not trivial [6], and even penetration of the aortic root by an Amplatzer device has been described [7].

Regardless of pulmonary vein morphology, interventional closure of an inferior sinus venosus defect must be ineffective, because the device cannot be anchored appropriately towards the IVC. Had the device been explanted earlier, damage to the mitral valve and the left atrium wall could have been prevented.
We therefore recommend that whenever there is doubt regarding the suitability of an ASD for interventional closure, a specialist for congenital heart disease is consulted, keeping in mind that ASD surgery can be performed through very limited skin incisions and with a very low complication rate [3]. If the result after percutaneous closure is not optimal, further diagnosis and surgical correction should be done early so as to avoid secondary complications. Ideally, performing such procedures in a modern hybrid operating room would allow for immediate surgical management should the intervention outcome be unsatisfying.

Conflict of interest: none declared.

REFERENCES