Embolization of the Device to the Left Pulmonary Artery after the Interventional Closure of Ruptured Sinus of Valsalva Aneurysm

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Formation of an aneurysm in the sinus of Valsalva of the aortic root is usually due to an area of congenital weakness in its wall. This aneurysm may progressively dilate and rupture into any of the cardiac chambers or into the pericardial cavity. Though this is conventionally treated by surgery, interventional therapy using various closure devices is becoming more common. Embolization of these closure devices may occur. We report a case of embolization of such a device into the left pulmonary artery which during surgical retrieval, unmasked the hidden ventricular septal defect (VSD). Therefore one has to be cautious while making a diagnosis of rupture of the sinus of Valsalva of right coronary sinus without VSD.

Key words: 1. Aneurysm  
2. Device  
3. Pulmonary artery  
4. Rupture

CASE REPORT

Rupture of the sinus of Valsalva (RSOV) is conventionally treated by surgery [1]. Interventionalists have recently experienced success in closing these defects using devices designed to close atrial septal defects or a patent ductus [2,3]. Several complications of this procedure, including embolization of the devices to the descending aorta, are well documented. We report a case of embolization of a device used to close a RSOV into the left pulmonary artery with concomitant unmasking of a hidden ventricular septal defect (VSD), which had not been identified before the procedure.

A 44-year-old man presented with complaints of dyspnoea and palpitation on exertion. An examination demonstrated cardiomegaly and a continuous murmur at the left third intercostal space. An electrocardiogram showed left ventricular hypertrophy, and echocardiography showed a dilated aortic root. A colour Doppler examination showed a high-velocity jet originating from the right coronary sinus (RCS) of the aorta and entering the right ventricular outflow tract (RVOT) (Fig. 1). When cardiac catheterisation was performed, the aortogram showed the contrast passing freely from the RCS into the RVOT. No VSDs were visualised, and the pulmonary artery pressure was normal.

A diagnosis of a RSOV aneurysm without VSD was made. An attempt was made to close the defect with a 16/14 size Lifetech patent ductus arteriosus closure device (Lifetech Scientific Co. Ltd., Shenzhen, China). After successful deployment, accurate positioning of the device was confirmed by an aortogram (Fig. 2) and an echocardiogram.
Three days after the procedure, the patient began to experience cough and dyspnoea. A chest X-ray (Fig. 3) and fluoroscopy (Fig. 4) revealed embolisation of the device into the left pulmonary artery. Fluoroscopy-guided percutaneous device retrieval was unsuccessful, and the patient was taken for emergency surgical retrieval of the displaced prosthesis.

At surgery, the pulmonary artery was noticed to be tense and dilated to twice the size of the aorta. The RCS was aneurysmal, had ruptured, and was prolapsing through a 15-mm VSD into the RVOT. The device was palpable, wedged in the left pulmonary artery just beyond the origin of the anterior artery branch. A pulmonary arteriotomy was performed and the device was retrieved.

The aneurysm was excised and both this defect and the VSD were closed using a patch of Dacron fabric. The patient recovered uneventfully from the operation and was discharged.
on the seventh postoperative day.

### DISCUSSION

The sinuses of Valsalva at the root of the aorta may become aneurysmal, forming thin-walled saccular or tubular outpouchings. Such defects are mostly congenital and comprise 0.15%–0.24% of congenital cardiac anomalies [4,5]. They are usually located in the right coronary sinus (75%–90%) or the noncoronary sinus (10%–25%), and rarely in the left coronary sinus [1]. These aneurysms have been shown to develop in a weak area between the aortic annulus fibrosa and media, which emerges due to the failure of the distal bulbar septum to fuse with the truncal ridges [5].

In addition to a congenital aetiology, these aneurysms may occur secondary to trauma, infective endocarditis, or tertiary syphilis. Iatrogenic injuries to the sinuses during VSD closure or during debridement of a calcified aortic or mitral valve may also result in aneurysm formation.

Sinus of Valsalva aneurysms (SVA) are often associated with other congenital defects, most commonly VSD and aortic regurgitation [1]. RCS aneurysms are more frequently associated with VSDs than non-coronary sinus aneurysms. Other anomalies are also known to co-occur with SVA, such as pulmonary stenosis, atrial septal defects, bicuspid aortic valve, patent ductus arteriosus, coarctation of the aorta, and subaortic stenosis [1,4].

The aneurysm and rupture of the coronary sinuses may occur at any age, with a male preponderance [1]. The aneurysmal pouch usually has an intra-cardiac course, but may protrude into the pericardial space. An aneurysm can simultaneously rupture into more than one site. Most RCS aneurysms
rupture into the right ventricle (73%), while most non-coronary sinus aneurysms rupture into the right atrium (86%) (Fig. 5) [1]. Aneurysms can rarely rupture into the left ventricle, left atrium, pulmonary artery, or the pericardial cavity.

Clinical symptoms of RSOV include the sudden onset of dyspnoea, chest pain, palpitation, fatigue, or orthopnoea due to a significant left-to-right shunt [4]. The rupture and its symptoms are generally precipitated by exertion. Echocardiography is the gold standard for the diagnosis of RSOV and the identification of other co-existing congenital anomalies [5]. Catheterisation is carried out in patients who require an evaluation of their coronary artery anatomy or if an interventional procedure is planned [1]. In this patient, a VSD was missed on an echocardiogram and on cardiac catheterisation, probably due to the large aneurysmal RCS that completely obstructed the VSD. Our patient underwent device closure since the VSD was not visualised. Therefore, one must be cautious when diagnosing RSOV of the RCS without VSD.

Patients with RSOV are best treated expeditiously [1]. Surgery is the conventional method of treatment, having a <4% mortality rate and good long-term results [4]. However, recurrences have been reported [3]. Non-ruptured SVAs requiring surgical intervention include those in patients with VSD, ventricular arrhythmia, infection, coronary ostial obstruction, RVOT obstruction, severe AR, and extra-cardiac SVA [4].

The era of catheter-based device closure of RSOV began in 1994 with Cullen et al. [3] successfully deploying a modified Rashkind umbrella device to close an RSOV. Since then, many other occluder devices have been utilised, with overwhelmingly favourable results [2]. Catheter-based procedures are currently considered to be the treatment of choice. Interventional procedures are particularly beneficial in the presence of previous sternotomy, recurrence, or severe co-morbidities [2].

The size of the device used for RSOV closure must be accurately assessed, since a large device may interfere with coronary blood flow or aortic valve cusp movement. However, a device of suboptimal size might dislodge and embolise, or result in a significant residual shunt.

Success rates up to 90% have been reported in catheter-based closures. Complications, although rare, include cardiac perforation, fistula formation, thrombosis, and device embolisation into the systemic or pulmonary circulation. These complications result in acute symptoms and haemodynamic compromise, requiring urgent surgical retrieval.

Post-deployment follow-up includes the assessment of coronary blood flow, aortic valve function, and the presence of thromboembolism. After the deployment of a device, a short course of anticoagulants or antiplatelet drugs is recommended to prevent thromboembolism until the endothelialisation of the device occurs [3]. The risk of infective endocarditis is unknown, and hence the need for life-long antibiotic prophylaxis is debatable [3].

In conclusion, RSOV, which is conventionally treated by surgery, is currently being successfully treated using occluder devices. As more centres take up such interventional procedures, varied complications are being reported. Despite the rapidly growing enthusiasm for the catheter-based device closure of RSOV, we should be aware of the fact that VSDs can be missed before the procedure. We report a case of the embolisation of a device used to close a RSOV into the left pulmonary artery.

CONFlict OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES