Percutaneous repair of an aortopulmonary fistula using platinum coils

Introduction

Preoperative investigations can sometimes reveal very abnormal and unexpected results. This case focusses on a previously unidentified aortopulmonary fistula in a well patient about to undergo cataract surgery. Despite deteriorating symptoms over previous years the patient was highly functioning and until more invasive investigation had been carried out the significance of the cardiac abnormality could not be delineated. This report describes the unusual presentation, the sequence of investigations and novel interventional procedure that led to closure of this patient's aortopulmonary fistula.

Discussion

Coronary artery fistulae are rare (0.05–0.2% of coronary angiography). Of these 15–20% are coronary to pulmonary artery fistulae; aorta to pulmonary artery fistulae are much more rare. Aorta to pulmonary artery fistulae can cause significant morbidity and are fatal if untreated (MacIntosh et al, 1991).

Aortopulmonary fistulas can be a primary abnormality (Kokotsakis et al, 2007), or develop as a consequence of previous thoracic surgery, especially to the descending thoracic aorta (Mukadam et al, 2005; Kokotsakis et al, 2007); trauma and infective processes such as tuberculosis are less common (Yoo et al, 2001). Symptomatic decline was most likely caused by the resulting pulmonary hypertension and atrial tachyarrhythmia.

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Aortopulmonary fistulae can present at any age, if congenital they are most commonly detected as a murmur in an otherwise asymptomatic child. If undetected or formed secondary to other pathology patients can become symptomatic at any age. Symptoms normally relate to cardiac insufficiency, e.g. dyspnoea on exertion, angina, fatigue, palpitations and even high-output congestive heart failure in patients with large fistulae (Liberthson et al, 1979).

This case is very unusual not only because of the origin of the ectopic vessel, but also the presentation of a refractory tachycardia with associated symptoms of dyspnoea and palpitations. This warranted further investigation.

A high index of suspicion is required to diagnose aorta to pulmonary artery fistulas. Transthoracic and transoesophageal echocardiography were very useful in this case to identify an ectopic vessel with abnormal flow on Doppler assessment. Despite being invasive cardiac catheterization remains the imaging modality of choice, providing not only the best diagnostic information but also the means for endovascular treatment. Cardiac magnetic resonance imaging is an improving method of identifying aorta to pulmonary artery fistulas and was used to determine the calibre of the vessel in this case (Weymann et al, 2009).

Traditionally closure of aorta to pulmonary artery fistulae has always been surgi-

Case Report

A 77-year-old woman admitted for cataract surgery was noted to have atrial tachycardia on a routine electrocardiograph. When referred to a cardiologist she gave a history of deteriorating exertional dyspnoea and palpitations over several years.

On examination the patient had a heart rate of 150 beats/minute with co-existing flow and regurgitant murmurs over the aortic valve, as well as mitral regurgitation, but no evidence of cardiac decompensation. There was a stable atrial flutter with a 2:1 block on the electrocardiograph, requiring large doses of amiodarone to return to sinus rhythm. The tachycardia was refractory to pharmacological treatment and the patient's deteriorating symptoms prompted further investigation.

Echocardiography identified mild mitral regurgitation with preserved left ventricular systolic function. A continuous shunt originating from the aorta with pulmonary hypertension was demonstrated by transoesophageal echocardiography.

Computed tomography scanning failed to identify any significant abnormality. Cardiac catheterization was required to visualize the abnormal flow seen on the transoesophageal echocardiogram (*Figure 1*). It showed an abnormal tortuous vessel originating superior to the native right coronary artery ostium and draining into a hugely dilated left pulmonary artery system. This caused significant pulmonary hypertension and was undoubtedly the cause of the patient's symptoms. Cardiac magnetic resonance imaging confirmed these findings and was used to estimate the calibre of the ectopic vessel (8.3 mm) before a therapeutic endovascular procedure.

The first attempted percutaneous closure, undertaken using an Amplatzer Vascular Plug, was abandoned because of the tortuosity of the fistula which had a corkscrew configuration, preventing plug delivery.

The second procedure used a Terumo Progreat microcatheter to sequentially deliver three Cordis Trufill platinum coil devices into the fistula (*Figures 2* and *3*). Within 10 minutes there was significant thrombosis and pro-grade flow reduction within the fistula. The right coronary artery and its ostium were unaffected. By the end of the procedure there was a 70% reduction in fistula flow with continuing thrombosis and no evidence of complications.

At 3-month clinical review, the patient described a marked improvement in her previous symptoms of disabling dyspnoea and greater than 90% improvement in walking capacity.

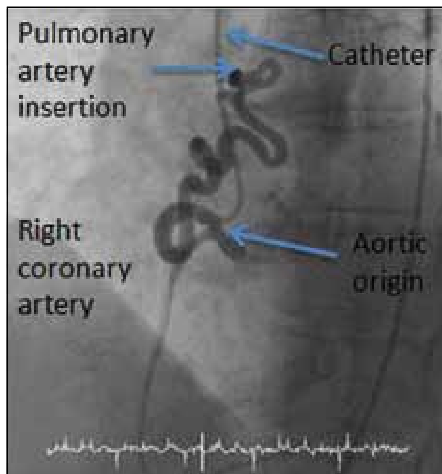

Figure 1. Catheterization of an aorto-pulmonary fistula to demonstrate configuration.

cal, until the use of endovascular repair was developed. Assessment before either surgical or endovascular repair by cardiac catheterization is essential to determine the haemodynamic significance of the fistula, its size, origin and point of insertion (Armsby et al, 2002).

Aorta to pulmonary artery fistulas rarely close spontaneously unless very small, but normally enlarge and require intervention. Closure is mandatory in large fistulas or if symptoms or complications are present. As their size increases there is not only symptomatic decline but increased risk of eventual rupture and endocarditis. Other complications of aorta to pulmonary artery fistulas include arrhythmias, intracardiac shunts, myocardial infarction, aneurysm, cardiac failure and even sudden death (Misra and Tandon, 2003).

There are multiple surgical and percutaneous transcatheter closure techniques described of fistulas, normally aortobronchial or coronary fistulas, but percutaneous closure of an aorta to pulmonary artery fistula is not published.

Conclusions

This case demonstrates that percutaneous platinum coil fistula occlusion is safe, feasible and effective despite proximity to other essential conduits. Aorta to pulmonary artery fistula closure is potentially more difficult, more complex and higher risk than coronary fistula. Nearly total occlusion was achieved and no re-study was required as the patient was clinically well and symptom free.

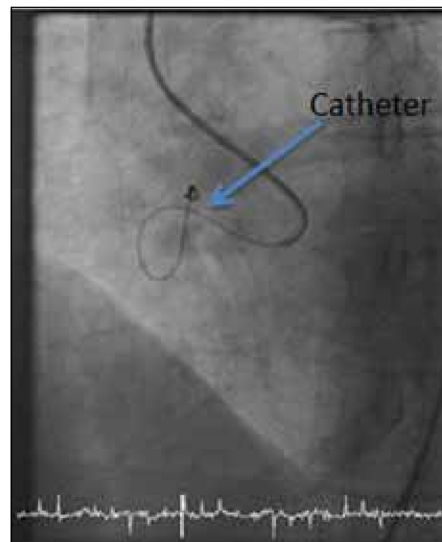

Figure 2. Delivery of first platinum coil into mid-segment of fistula.

There are no long-term or trial data of this technique but evidence of the feasibility, symptomatic relief and short–mid-term success is seen in this patient. **BJHM**

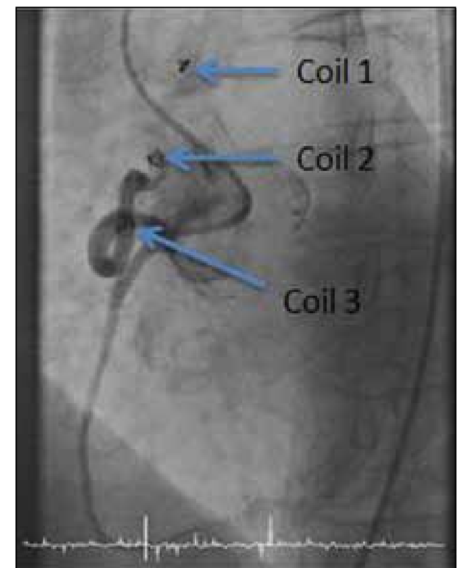

Figure 3. Reduction in fistula flow after delivery of three platinum coils.

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LEARNING POINTS

- This article presents a highly unusual case of an aortopulmonary fistula in an adult, highlighting the need for a high index of suspicion to aid diagnosis.
- Aortopulmonary fistulas can be congenital or acquired as a result of previous thoracic surgery.
- Aortopulmonary fistulas generally become more symptomatic over time and can lead to life-threatening complications.
- While cardiac catheterization remains the imaging modality of choice, there is an increasing role for cardiac magnetic resonance imaging in detecting structural cardiac abnormalities.
- In this case percutaneous intervention obviated the need for open operative ligation and was clearly beneficial as an alternative to surgery in an elderly patient.