We present a case of a 23-year-old man with Shone’s complex, including bicuspid aortic valve, coarctation of the aorta status post neonatal repair and subsequent stent placement, mitral valve stenosis status post valve repair, and eventual replacement 1 year before the current presentation. A noncontrast thoracic computed tomography scan was performed after his mitral valve replacement to investigate a persistent pleural effusion. The scan revealed an unusual area of high and low attenuation within the proximal pulmonary arterial system, and a contrast-enhanced computed tomography study was recommended for additional evaluation. However, given that the patient was asymptomatic with unremarkable physical examination, follow-up study was not performed until ≈1 year later when he was noted with a new continuous murmur on physical examination and an echocardiogram revealed continuous flow arising from the distal ascending aorta into the pulmonary artery on two-dimensional color Doppler (Figures 1A and 2, arrow; see Movies I through III in the online-only Data Supplement) and continuous-wave Doppler (Figure 1B), although direct visualization of the pulmonary arteries was limited because of acoustic windows. Given the presence of the aortic stent with potential for susceptibility artifact obscuring the pulmonary artery with magnetic resonance imaging, a contrast-enhanced computed tomography was performed and revealed a small aortopulmonary window (APW) between the ascending aorta and main pulmonary artery (Figure 3A and 3B, arrow), as well as an associated dissection involving the main and left pulmonary arteries (Figure 3C and 3D, arrowheads). Given the location and timing of presentation, it was presumed that this acquired APW and pulmonary artery dissection were related to aortic cannulation and the aortic cross clamp during the mitral valve replacement. The APW was successfully closed percutaneously using a 5-mm Amplatzer Septal Occluder device without complication (Figure 4; Movies IV and V in the online-only Data Supplement). Follow-up computed tomography showed stable position of the occluder device (Figure 5A and 5B, arrow) and persistent, although decreasing, size of the pulmonary artery false lumen (Figure 5A through 5D, asterisk). Our patient had 2 unusual findings as a result of aortic cannulation: (1) APW and (2) pulmonary artery dissection. Aortic cannulation has been associated with the development of aortopulmonary fistulae and aortic aneurysms. There have been multiple reports of acquired APW as a result of balloon pulmonary angioplasty. Another unusual aspect of our case is the presence of pulmonary artery dissection, a rare finding in the absence of pulmonary hypertension that is typically fatal and often diagnosed at autopsy. There has been 1 case of congenital APW with pulmonary artery dissection and rupture related to chronic pulmonary hypertension, but to our knowledge, there have been no reports of APW or pulmonary artery dissection related to aortic cannulation.

We believe that the APW in our patient was a result of traumatic aortic cannulation during mitral valve replacement with direct iatrogenic injury to the pulmonary artery wall with resultant development of APW and pulmonary artery dissection at the time of surgery. Our patient remained asymptomatic for >1 year despite these complications and underwent successful percutaneous closure of his APW. This case highlights an unusual complication of aortic cannulation and demonstrates that patients may remain asymptomatic despite having 2 potentially fatal surgical complications.

Disclosures

None.

References


From the Department of Pediatrics, Division of Pediatric Cardiology, New York University School of Medicine, New York, NY (R.F.A., M.A.); and Department of Medicine, Division of Cardiology, Medstar Georgetown University Hospital, Washington, DC (M.B.S.). The online-only Data Supplement is available at http://circ.ahajournals.org/lookup/suppl/doi:10.1161/CIRCULATIONAHA.112.001364/-/DC1. Correspondence to Robert F. Adams, MD, 13421 N 1st Ave, Boise, ID 83714. E-mail RobertFAdamsMD@gmail.com (Circulation. 2013;128:e180–e181)

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Figure 1. A. Suprasternal notch echocardiogram showing turbulent shunt arising from the distal ascending aorta into the pulmonary artery (arrow). B. Continuous-wave Doppler demonstrating continuous shunting in systole and diastole consistent with an aortopulmonary connection.

Figure 2. Parasternal short axis echocardiogram at the level of the aortic valve (A) with color Doppler images in systole (B) and diastole (C) demonstrate turbulent flow in the main pulmonary artery (arrow) consistent with aortopulmonary connection. Ao indicates aorta; and MPA, main pulmonary artery.

Figure 3. Multiplanar reformatted contrast-enhanced computed tomographic angiography images (A through C) and 3-dimensional volume-rendered image (D) demonstrating aortopulmonary connection (arrows, A and B) and corresponding pulmonary artery dissection (arrowheads, C and D).

Figure 4. Angiography with an aortic root injection demonstrating the Amplatzer Septal Occluder device (arrow) in position within the aortopulmonary window with no residual shunt.

Figure 5. Multiplanar reformatted contrast-enhanced computed tomographic angiography images (A through C) and 3-dimensional volume-rendered image (D) demonstrating the Amplatzer Septal Occluder device (arrows, A and B) and persistent but decreasing size of pulmonary artery dissection (asterisks, B and D).
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