

Mycotic Pseudoaneurysm Associated with Aortic Coarctation

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ABSTRACT

Development of a mycotic aneurysm or pseudoaneurysm after subacute bacterial endarteritis is uncommon. Nonetheless, patients with coarctation of the aorta are more likely to develop this complication. We describe a case of a large pseudoaneurysm discovered in a child with a previously undiagnosed aorta coarctation. Successful repair was performed with the aid of partial left heart bypass and the use of an interposition graft. A high index of suspicion is necessary to accurately diagnose this rare but life-threatening entity.

KEY WORDS: Coarctation; Diagnosis; Mycotic aneurysm; Surgery.

CASE REPORT

A 2 1/2-year-old previously healthy boy experienced approximately 5 months of malaise, weight loss, and intermittent low-grade fevers. He had been recently treated for sinusitis with an associated respiratory infection. On a car trip with his parents, he experienced severe lower abdominal and lower leg pain and was inconsolable and unable to find a comfortable position.

Evaluation by his primary care physician included a chest x-ray that revealed moderate cardiomegaly and prompted referral to a pediatric cardiologist. His physical examination, chest x-ray, echocardiogram, and chest computed tomogram were consistent with an aortic pseu-

doaneurysm at the site of a previously undiagnosed aortic coarctation. He was electively intubated and started on an Esmolol drip at 250 µg/kg/min. Blood cultures were positive for streptococcal sanguis, and he was treated with intravenous penicillin and gentamicin. He was transferred to the University of Michigan Medical Center for surgical intervention.

Physical examination revealed a pale, thin, ill-appearing 2 1/2-year-old boy. His weight was 12.3 kg, heart rate 130 beats/min, blood pressure in the right upper extremity 160/70 and left lower extremity 75/55, with a respiratory rate of 20 controlled by mechanical ventilation. His lungs were well aerated with no evidence of rhonchi or rales. His cardiac examination showed a hy-

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perdynamic apical impulse that was displaced to the anterior axillary line. The first and second heart sounds were normal. A III/VI midsystolic murmur was audible and heard best at the left upper sternal border with radiation to the axilla and continuation into diastole. An S_3 gallop was also audible. His abdomen was soft, the liver edge was palpable at 2.5 cm below the right costal margin, and a spleen tip was palpable in the left upper quadrant. No other abdominal masses were noted. His upper extremities were warm with accentuated pulses, whereas his lower extremities were cool with delayed and diminished femoral pulses. Laboratory evaluation revealed normal electrolytes with a blood urea nitrogen of 21 mg/dl and a creatinine of 0.5 mg/dl. His white blood cell count was $13,000/\text{mm}^3$ with 63% neutrophils, 29% lymphocytes, 7% monocytes, and 1% eosinophils. Urinalysis showed a specific gravity of 1.020, 10–25 red blood cells, and 3–5 white blood cells per high power field. An electrocardiogram revealed normal sinus rhythm with a rate of 123 beats/min, right atrial enlargement, and borderline left ventricular hypertrophy. A chest x-ray revealed moderate cardiomegaly, a mass in the left upper lung field consistent with an aneurysm of the descending thoracic aorta, and mild pulmonary parenchymal cephalization (Fig. 1). An echocardiogram revealed a dilated poorly contractile left ventricle with a shortening fraction of 11% and two-dimensional ejection fraction of 20%. A juxtaductal coarctation was noted with a peak velocity of 4 m/sec.

To better delineate the anatomy of the proximal aortic arch and the extent of the pseudoaneurysm, a magnetic resonance imaging (MRI) study was performed. Images were acquired on a 1.5-T magnet (Signa, General Electric Medical Systems, Milwaukee, WI). The thoracic MRI was performed using "black blood" spin echo (Fig. 2), "white blood" phase contrast (Fig. 3), and gadolinium-enhanced three-dimensional volume MR angiography techniques (Fig. 4). A bi-lobed extraluminal collection was identified surrounding the proximal descending thoracic aorta, demonstrating swirling dephasing artifact due to turbulent flow. Three-dimensional gadolinium-enhanced images clearly showed the necks of a pseudoaneurysm that arose from the anterior and left lateral surface of the aorta approximately 4 cm from the origin of the left subclavian artery. The posterior component of the dumbbell-shaped pseudoaneurysm measured 7.5×6.5 cm with the smaller anterior component measuring 3.5×2.0 cm.

The patient underwent surgical repair via a left posterolateral thoracotomy using partial left heart bypass

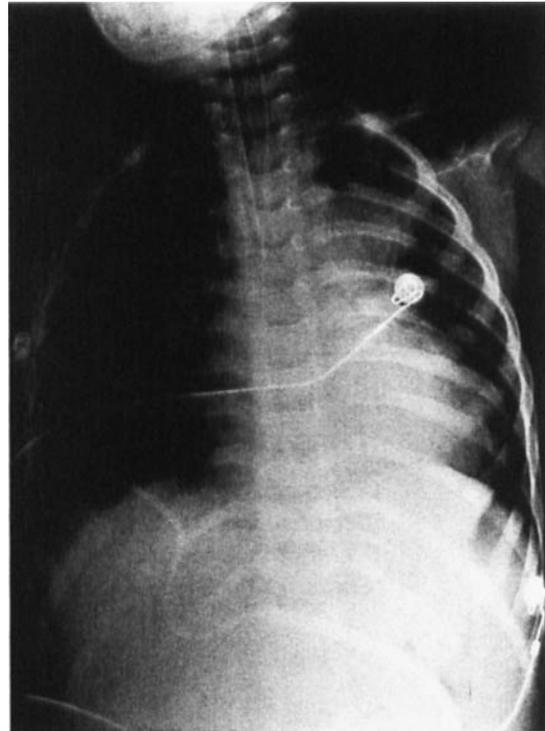


Figure 1. Frontal chest radiograph demonstrates moderate cardiomegaly, an abnormal superior mediastinal contour with focal ill-defined rounded opacity projecting at the level of the aortopulmonary window.

(left atrial to descending thoracic aorta). A large dumbbell-shaped pseudoaneurysm was found eroding into the mediastinum and left pleural space and taking origin from two distinct sites of rupture just distal to the coarctation site. A total of 5 cm of aorta was involved in this inflammatory process, beginning at the base of the left subclavian artery. There was evidence of dissection in the wall of the aorta just distal to the coarctation with compression of the true aortic lumen by the pseudoaneurysm. Proximal and distal vascular control was obtained, the aorta was transected in areas of normal tissue, the mediastinal portion of the pseudoaneurysm was left in situ, and continuity was restored with a 12-mm Hemasfield interposition graft.

Culture of the resected aortic tissue revealed no viable organisms. Microscopic examination of the resected aorta revealed disruption of the elastic lamina and acute and chronic inflammation. The boy's convalescence was uncomplicated. He was discharged 6 days after surgery to receive a total of 6 weeks of intravenous antibiotics.

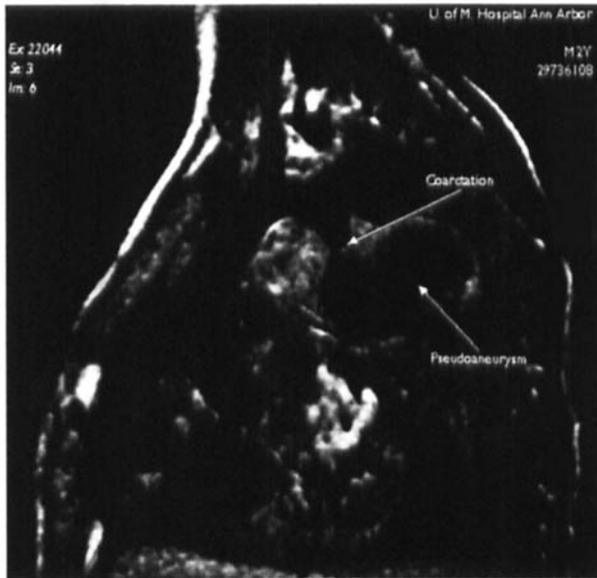


Figure 2. Sagittal, oblique, spin echo, T1-weighted (TR 618/TE 20, 2 NEX) image demonstrating level of coarctation and large adjacent pseudoaneurysm with flow void.



Figure 4. Sagittal, oblique, maximum intensity projection reconstructed image from three-dimensional gadolinium-enhanced MR angiogram demonstrates relation of large bilobed pseudoaneurysm to origin of great vessels and site of coarctation.



Figure 3. Gadolinium-enhanced, sagittal, oblique, gradient recalled, cine-MR angiogram, phase image (fast multiplanar gradient recalled echo sequence TR 10.2/TE 3.1, flip angle 30 degrees, 4 NEX) demonstrates decreased signal in the pseudoaneurysm due to turbulent flow dephasing artifact.

DISCUSSION

The natural history of patients with coarctation of the aorta was clearly outlined by Abbott in 1928 (1) and later reviewed by Reifenshtein in 1947 (2). Reported complications associated with coarctation include hypertension, congestive heart failure, cerebrovascular accidents, bacterial endarteritis, aortic aneurysms and pseudoaneurysms, and aortic rupture (3–5). Aortitis accounted for approximately 20% of deaths in these series, and before operative intervention, an 18% incidence of aortic rupture was reported in patients with coarctation of the aorta.

Aortitis in the setting of an aortic coarctation is the result of infected emboli that invade the elastic and muscular components of the vessel wall. Rupture may occur in the thoracic aorta above the coarctation or more commonly in the area of poststenotic dilatation in the descending thoracic aorta (6). Abbott believed that the mycotic aneurysm tended to be localized to the aorta distal to the coarctation due to irregularities in the lumen, leading to turbulent flow and providing a nidus for bacteria growth (1,5,7), and termed this “locus minoris resistentiae.” The infecting bacterial organisms were most often

Streptococcus species, although *Pneumococcus* and *Staphylococcus aureus* were noted on occasion. These aneurysms characteristically occurred in young boys between the ages of 10 and 20 years, although a few reports described cases in the first few years of life (8,9). The mortality rate of a mycotic aneurysm associated with coarctation of the aorta if left untreated approaches 100% and is most often a result of rupture of the aneurysm into the esophagus, bronchus, or pleural cavity.

Although many reports exist of mycotic aneurysms complicating surgical repair of a known coarctation, a detailed history, physical examination, and high index of suspicion is necessary to diagnose aortitis in association without known aortic pathology. Schneider et al. (10) reported that fever, hematuria, and diminished femoral pulses were a highly sensitive triad, suggestive of coarctation of the thoracic aorta complicated by bacterial endarteritis. Oldham et al. (11) similarly reported on 13 patients exhibiting this rare complication of mycotic aneurysm associated with coarctation of the aorta. Their diagnosis was established by a combination of febrile illness, positive blood cultures, and development of a mediastinal mass in a patient with clinical findings of aortic coarctation. They cautioned that the average time interval between a normal chest film and one demonstrating the aneurysm was 7 weeks but ranged from as little as 6 days to 4 months.

The first successful treatment of a mycotic pseudoaneurysm complicating an aortic coarctation was reported by Schumacker et al. in 1948 (12). In Oldham et al.'s review of 13 cases (11), 10 patients survived repair using a combination of primary reanastomosis and a variety of interposition grafts.

Treatment of a newly discovered pseudoaneurysm of the thoracic aorta associated with endarteritis at a coarctation site should be prompt. Multiple blood cultures should be drawn and the patient begun on the appropriate intravenous antibiotics and stabilized. The systemic blood pressure should be controlled with intravenous beta-blockers (Esmolol) in an attempt to decrease the rate and force of left ventricular ejection against the wall of the pseudoaneurysm. Imaging studies such as computed tomography of the chest or MRI will clearly define the extent of the aneurysm and determine the optimal surgical approach. Surgical repair is thus then best performed

using partial left heart bypass to lower the thoracic aortic pressure while maintaining cerebral and distal perfusion. Usually, an interposition graft will be required because there is generally considerable destruction of the aortic wall. Intravenous antibiotic therapy should be continued for approximately 6 weeks after the repair. If performed on an elective basis before erosion into mediastinal or pleural structures or rupture leading major hemorrhage, a successful outcome can be achieved.

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