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Late Development of Aortic Pseudoaneurysm after Coarctation Repair with Fistulization to the Bronchial Tree. A Case Report

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ABSTRACT

Background: Fistulous communication between the aorta and the tracheobronchial tree is an uncommon and serious cause of hemoptysis secondary to complications of a previous operation performed on the aorta. In cases in which an appropriate surgical intervention is carried out, the survival rate approaches 76%. This surgery is considered one of the most risky operations on the aorta, challenging the surgeon's ability to resolve the problem.

Methods: We present the case report of a 43-year-old female with massive hemoptysis. Her medical history disclosed repair of coarctation of the aorta (15 years before). She underwent emergency left thoracotomy; surgical exploration revealed a false aneurysm from the previous aortic patch repair which communicated to a subsegmental bronchus of the left upper lobe.

Results: The thoracic aorta was isolated and clamped, and the previous patch was removed. The bronchial side of the fistula was managed with left superior lobectomy and the aorta was repaired with the placement of a coated woven dacron graft onto healthy aortic tissue.

Conclusions: The patient had an uneventful recovery and remains asymptomatic six months after discharge.

INTRODUCTION

Aneurysm formation following coarctation repair has been seen in patients who have had patch aortoplasty using prosthetic material. It is theorized that differential compliance between the prosthetic patch and the native aorta opposite the patch is one of the causes of these aneurysms [Christensen 1948, Del Nido 1986, Luosto 1980, Bromberg 1989]. In 1947, Jones reported the first successful surgical repair of an aortobronchial fistula in a patient that had previously undergone patent ductus arteriosus ligation [Jones 1947]. Massive hemoptysis tends to be the final episode of aortic fistulization into the tracheobronchial tree [Julia-Serda 1996]. Before reaching this stage the patient typically has self-limiting episodes of hemoptysis of a lesser intensity. This presentation is characteristic and is attributed to the thrombotic obstruction of the fistula and its opening, due either to the lysis of the thrombus or to the enlargement of the communication. This is why an adequate clinical evaluation, thorough survey of medical history (such as previous aortic interventions) and x-ray findings (such as widening of the mediastinum) are crucial in reaching a diagnosis as early as possible [Demeter 1980, Graeber 1980]. Thoracic CT scan and aortography are the imaging methods in the diagnosis of aortic aneurysm [Posniak 1989]. Aortobronchial fistulae, if left untreated, are almost always fatal; however, in cases in which an appropriate surgical intervention is carried out, survival rates reach 76%. Nevertheless, the real incidence and surgical risk of thoracic aortic aneurysm associated with a bronchial fistula are difficult to ascertain since many cases may be unreported [Barbash 1983, Riancho 1989, McIntosh 1991].

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CASE REPORT

A 43-year-old female was admitted to our institution with massive hemoptysis. Her medical history disclosed repair of coarctation of the aorta fifteen years before. The patient was asymptomatic until 3 weeks before admission, when she complained of "spitting up blood". The source of the bleeding could not be further characterized by history. The patient expectorated several hundred milliliters of blood on three occasions. On arrival the pulse was 89 beats/min, respiratory rate 19 breaths/min, and BP 110/70 mm Hg. Examination revealed diffuse rales over both lungs. Cardiac examination, EKG, and routine labs were normal. A chest x-ray revealed a large, smooth mass posterior to the aortic knob which had the appearance of an aortic aneurysm (see Figure 1 ◎). CT scan showed a large left thoracic mass involving the left superior pulmonary lobe and a segment of descending thoracic aorta, with severe intrapulmonary hemorrhage (see Figure 2 ◎). Over the

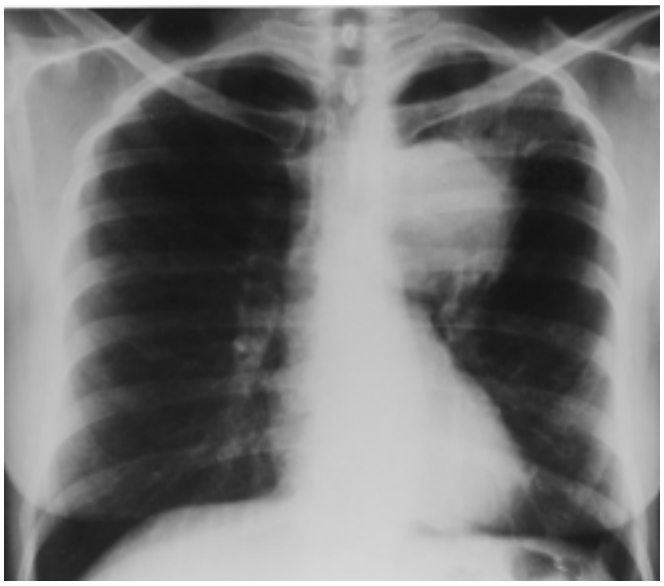

Figure 1. Preop. PA Chest x-ray

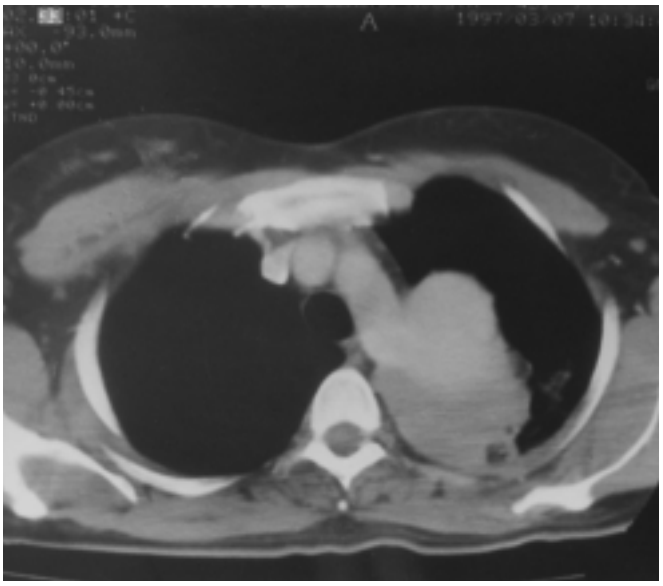

Figure 2. Preop. Chest CT scan

following eight hours her respiratory distress and hypoxemia progressed, necessitating endotracheal intubation. The Department of Cardiovascular Surgery was consulted; upon our arrival the patient was quickly evaluated and immediately transferred to the operating room for emergency thoracotomy. A left lateral thoracotomy was performed revealing the lung densely adherent to the parietal pleura. Exploration revealed a false aneurysm from the suture line of the previous aortic patch repair communicating with a subsegmental branch of the superior bronchus, and bloody infiltration of the entire lobe. The thoracic aorta was isolated and clamped proximally below the subclavian artery and distally at its mid-descending portion. Removal of the aneurysmal tissue, prosthetic material, and diseased aortic tissue was accomplished. Vascular repair required placement of a 20-mm coated woven dacron graft. Finally, a left upper lobectomy was performed. The patient had an uneventful recovery. Microscopic evaluation of the specimen demonstrated adventicial infiltrates composed primarily of lymphocytes with few plasma cells and histiocytes. The media showed considerable atrophy associated with intimal plaque and adventicial inflammation. After a six-month follow-up period, echocardiogram remains normal and the patient asymptomatic and fully rehabilitated.

DISCUSSION

The majority (87%) of reported cases of aortobronchial fistulae involve an aneurysm of the descending thoracic aorta. Enlargement of the aneurysm causes tracheobronchial compression and its adherence to surrounding structures, leading to chronic inflammation, infection, foreign body reaction, or pressure necrosis. Rupture can be confined, open to the tracheobronchial tree or free into the pleural space. The incidence of thoracic aortic aneurysm is similar for men and women; however, more than two thirds of the documented cases of aortobronchial fistulae have occurred in male patients. Aortic fistulization to the bronchial tree can develop in any decade, but the incidence generally increases with age. The clinical course of aortobronchial fistula is characterized by recurrent hemoptysis, with virtually all patients experiencing at least one episode. Massive hemoptysis, defined as the expectoration of more than 400 ml of blood in 24 hours, is reported to occur in more than half of the patients and is often the terminal event. Graeber and associates stated that plain chest x-ray films are the most helpful diagnostic study because of their representation of lung abnormalities adjacent to the aneurysm [Graeber 1980]. However, an aneurysm was diagnosed in only seven of 44 cases when these chest x-ray abnormalities were present. Computed tomography is the method of choice in differentiating a mediastinal or paramediastinal lung mass from an aortic aneurysm [Graeber 1980]. The keystones to management include resuscitation, diagnosis, and definitive surgical intervention. Massive, persistent bleeding can be controlled immediately by intubation of the contralateral bronchus or placement of a balloon embolectomy catheter on the side of the hemorrhage. If these measures fail, emergency thoracotomy may be lifesaving as in the present case. Surgical treatment of aortobronchial fistula consists of the repair of both the aortic and pulmonary defects. Circulation distal to the fistula has been maintained by patch closure, direct suture, or prosthetic graft (usually the latter).

When a previously placed prosthesis is present, that graft should be removed because of the possibility of infection. The bronchial side of the fistula can be managed with resection (wedge, segmentectomy, lobectomy, pneumonectomy) or simple closure. Most commonly, pleural or pericardial flaps have been used to cover suture lines; others have used intercostal muscle to buttress the bronchial closure. Omental patch has been used successfully in the

presence of chronic mediastinal or thoracic infection involving a vascular prosthesis. Complete enclosure of the graft with viable tissue has potential benefits in terms of minimizing residual infection and should prevent recurrence of the aneurysm. The use of an extrathoracic skeletal muscle for this purpose rather than omentum avoids the added risk of opening a second body cavity in the presence of infection in patients who are already desperately ill. Without surgical intervention, aortobronchial fistula is uniformly fatal. Morbidity is substantial in this group of patients and commonly consists of pulmonary infection. The majority of these patients require prolonged post-op endotracheal mechanical ventilation; but with appropriate surgical treatment, most patients will make an excellent recovery. A high index of suspicion is necessary in making the diagnosis of aortobronchopulmonary fistula. As in the case described, the diagnosis should be considered in any patient experiencing minor or major hemoptysis with either coexisting thoracic aortic aneurysm or a history of thoracic aortic operation. CT scan is the best single diagnostic modality available; however, an aggressive surgical attitude for early intervention should be maintained.

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